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Case Report

Onset of Chronic Expanding Hematoma 25 Years After Total Hip Arthroplasty

Yugen Ogata, MD, Kosuke Kumagai, MD, PhD *, Tomohiro Mimura, MD, PhD, Shunichi Miyahara, MD, Masashi Egawa, MD, Hideki Saito, MD, Yasutaka Amano, MD, Takafumi Yayama, MD, PhD, Mitsuhiko Kubo, MD, PhD, Shinji Imai, MD, PhD

Department of Orthopaedic Surgery, Shiga University of Medical Science, Otsu, Japan

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ABSTRACT

Chronic expanding hematoma (CEH) is a rare anatomical condition that gradually expands due to trauma or surgery. We report the case of a 56-year-old woman who developed CEH 25 years after metal-on-polyethylene total hip arthroplasty. She presented with swelling and radiating pain in the right inguinal region. Tocilizumab was administered for treating rheumatoid arthritis and renal amyloid A amyloidosis. Diagnostic imaging and partial resection revealed a soft tissue mass and a CEH, respectively. The symptoms recurred 6 months later; dialysis was initiated, and the CEH was resected under general anesthesia, leading to improvement. This case report emphasizes the importance of prompt diagnosis and intervention in CEH management for preventing further complications and improving the patient's quality of life.

Introduction

Soft-tissue masses around the site of total hip arthroplasty (THA) have been well documented [1-3]. They most commonly arise as pseudotumors owing to a granulomatous reaction to particulate debris generated by the THA prosthesis. Pseudotumors may be associated with osteolysis [4] and, more rarely, with chronic expanding hematomas (CEHs); thus far, only 3 cases have been reported after THA. Two of these cases were reported after metal-on-metal (MoM) THA [5,6]; the second case was reported to have occurred after ceramic-on-polyethylene (CoP) THA [7]. MoM THA is associated with tumor-like cystic lesions; therefore, MoM THA and CEH may be correlated. However, only one case of CEH has been reported, and the hematoma developed more than 20 years after CoP THA was performed. To the best of our knowledge, we have presented the first case of bilateral CEH that developed after a long period following THA (ie, 25 years).

Case history

A 56-year-old woman initially presented to our institution 15 years after undergoing THA. She had bilateral hip deformities caused by rheumatoid arthritis (RA) mutilans. At a different hospital, she had undergone bilateral cementless THA with a conventional polyethylene cup at 30 years of age (CP socket; Kyocera Corporation, Kyoto, Japan). She also presented with renal amyloid A amyloidosis. Tocilizumab was administered for treating RA and amyloid A amyloidosis [8,9]. However, the patient complained of swelling and pain in the right inguinal region 10 years after the first consultation at our hospital (ie, 25 years after THA). Moreover, an elastic soft mass was found in the right inguinal region (Fig. 1), and pain was noted to radiate from the site. There was no associated heat or redness.

Blood chemistry findings revealed mild anemia, elevated inflammatory markers, and abnormal levels of coagulation factors (Table 1). A plain radiograph of the hip showed osteolysis in the greater and lesser trochanters (Fig. 2c). Considering the possibility of an implant infection and that an inflammatory reaction might have been masked by tocilizumab, arthrocentesis was performed. However, the joint aspiration fluid was blood-like and not purulent; furthermore, a bacterial culture analysis of the fluid yielded negative results. Therefore, we ruled out the possibility of infection in this case.

* Corresponding author. Department of Orthopaedic Surgery, Shiga University of Medical Science, Seta Tsukinowa-cho, Otsu, Shiga 520-2192, Japan. Tel.: +81 77 548 2252.
E-mail address: kumamp@belle.shiga-med.ac.jp

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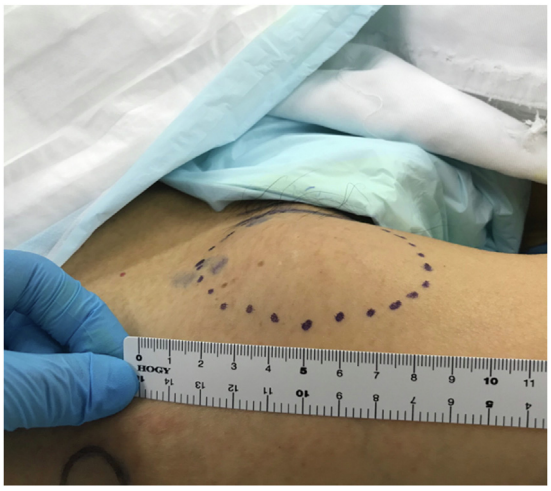

Figure 1. A mass of approximately 8 × 6 cm is observed in the right inguinal region. The mass is not associated with heat or redness, although it is accompanied by spontaneous pain.

We reviewed the patient's radiographs obtained over the course of 10 years following her first visit to our institution. No abnormal findings were found in the first 7 years (Fig. 2a). However, images (Fig. 2b) taken 7 years later (ie, 22 years after THA) revealed mild osteolysis inside the femoral neck in both hip joints (the left side is not shown in the figure).

Computed tomography confirmed the presence of a soft tissue mass with osteolysis in the bilateral hip joints, which was suggestive of either a reactive pseudotumor or the formation of an RA pannus (Fig. 3a and b). A radiologist diagnosed the pseudotumor to be reactive to the implant. Magnetic resonance imaging (MRI) revealed a soft tissue shadow protruding from the right hip joint to the right inguinal region (Fig. 3c-f). In fat-suppressed T2-weighted images, a low signal intensity was seen in the mottled region and margin of the mass (Fig. 3c-e). In a T1-weighted image, the signal was similar to that of the muscles, with some high signal areas (Fig. 3f). Based on these findings, humoral components, such as joint fluid, hematomas, pigmented villonodular synovitis, and synovial hemangiomas, were included in the differential diagnoses. The radiating pain was suspected to be due to compression of a femoral nerve branch.

We chose to perform a biopsy with partial resection under local anesthesia because general anesthesia could worsen the patient's renal function. During the surgery, a skin incision of approximately 5 cm was made directly above the mass. Furthermore, the nerve running through it was identified. This confirmed our theory that the pain was induced by compression of a femoral nerve branch. Active bleeding occurred after a capsule surrounding the mass was pierced. No pathological findings indicating amyloid and foreign body reactions (suggestive of metal or polyethylene usage) were noted. Furthermore, no lymphocyte aggregation characteristic of adverse reactions to metal debris was observed. Therefore, a diagnosis of CEH was made. After surgery, the symptoms resolved probably due to a reduced pressure on the nerve. However, the patient's renal function worsened 6 months later, and the pain in the right hip joint recurred. Therefore, we decided to remove the hematoma under general anesthesia after initiating dialysis. The right hip joint was filled with fresh and clotted blood; the hematoma was removed together with the joint capsule (Fig. 4a). Though the femoral implant was partially exposed in the femoral neck, there was no obvious loosening. The intraoperative bleeding volume was approximately 2300 mL, and the resected hematoma weighed 184 g (Fig. 4b). Postoperative pathological examination revealed only bloody components (Fig. 4c and d, #1–3). Regarding the macroscopic appearance of the resected hematoma, hematoxylin and eosin staining revealed component #1 to be a fresh hematoma, with lesser fibrinization than that observed in components #2 and #3. These findings were consistent with the features of a CEH. Because no metal powder was detected in the macrophages, metallosis was not suggested. After surgery, she recovered and could walk with the aid of a walker. At the 6-month postoperative mark, blood tests revealed no progression of anemia, and radiographs showed no spread of osteolysis. However, only 6 months have passed since the operation, and further follow-up is necessary. The patient provided informed consent and publication approval after she was informed that data concerning her case would be submitted for publication.

Discussion

CEH was first reported by Friedlander in 1968 [10]. In 1980, Reid defined it as a hematoma that gradually increased in size for over a month or more in the absence of an identified site of origin [11]. The mechanism underlying the development of CEH is unclear. Its size is assumed to increase due to repeated bleeding and resultant fibrosis from small blood vessels under the fibrous cap [11]. Labadie speculated that the blood and associated debris cause chronic inflammation. This in turn could stimulate the formation of new blood vessels in the capsule and cause further bleeding [12]. Dynamic computed tomography may indicate tumor margin enhancement during the arterial phase [13], whereas T2-weighted MRI may show a "mosaic pattern" (a mixture of fresh and old blood) [14]. However, these findings are nonspecific, and it is difficult to distinguish hematomas from malignant soft tissue tumors based on clinical and radiological features [15]. Therefore, a histopathological diagnosis based on the excised specimen is necessary. The histopathological features of a CEH include the following: a central region of blood walled off by an inner lining of granulation tissue and an outer edge of dense fibrous tissue [11]. In our case, we observed a relatively fresh hematoma and fibrinolysis around it, which were consistent with the findings of a CEH. In our case, imaging with a

Table 1
Blood panel results, including coagulation profiles and inflammation markers.

White blood cell (/μL)	8200	Prothrombin time (s)	11.9
Red blood cell (/μL)	3.57×10^6	Prothrombin time-international normalized ratio	1.03
Hemoglobin (g/dL)	9.9	Activated partial thromboplastin time (s)	27.0
Platelets (10^4/μL)	10.1	Fibrinogen (mg/dL)	238
Erythrocyte sedimentation rate: 1h (mm)	9.0	Blood coagulation factor XIII (%)	62
C-reactive protein (mg/dL)	0.06		
Procalcitonin (ng/mL)	2.01		
Creatinine (mg/dL)	3.49		
blood urea nitrogen (mg/dL)	94		
estimated glomerular filtration rate (mL/min/1.73 m^2)	11.5		

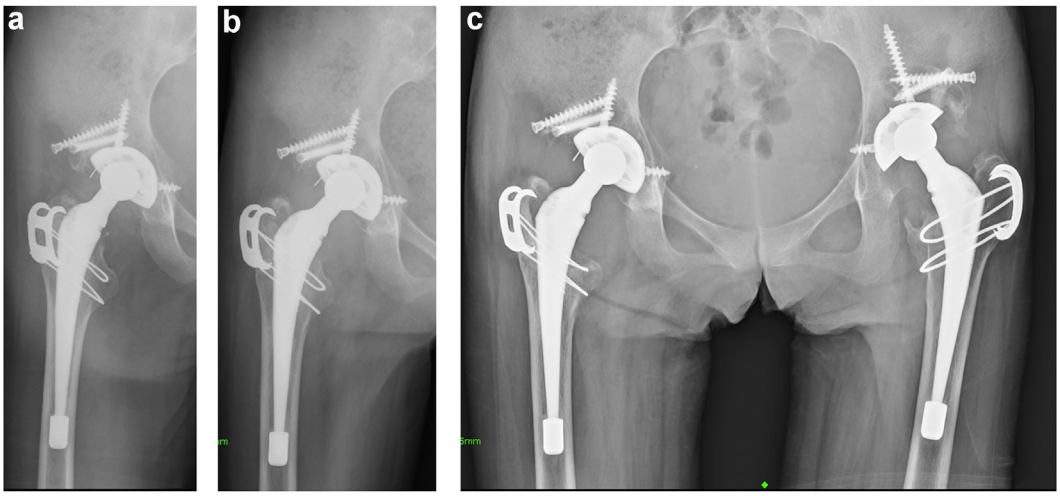

Figure 2. Radiographs showing the progression of osteolysis in the right hip over a 10-year period. (a) Anteroposterior (AP) view of the right hip obtained 10 years ago (15 years after THA) at the first visit to our hospital. No apparent osteolysis is seen. (b) AP view of the right hip obtained 3 years ago (22 years after THA) shows mild osteolysis around the lesser trochanter. (c) AP view of the pelvis obtained at the most recent consultation (25 years after THA) shows abnormal soft tissue shadowing in the right hip joint and extensive osteolysis in the greater trochanter and the upper half of the lesser trochanter on the right side (yellow arrow). In addition, in the left hip joint, bone resorption is observed not only in the femur but also in the acetabular side (red arrow).

contrast medium was not possible due to renal dysfunction, which made establishing an image-based diagnosis of a CEH difficult. A pseudotumor can occur around a hip prosthesis due to adverse reactions to the metal of the prosthesis or wear and corrosion of its head-neck interface [16]. Based on MRI, the pseudotumor (as seen in our case) showed a relatively uniform cystic pattern rather than a mosaic pattern [17,18]. It was unclear whether MoP played a role in the formation of the hematoma in our case. Amyloidosis may be involved in osteolysis [19], but no amyloid component was found in the hematoma.

Complete surgical resection of the hematoma, including the capsule, is considered the gold standard treatment for CEH. This is

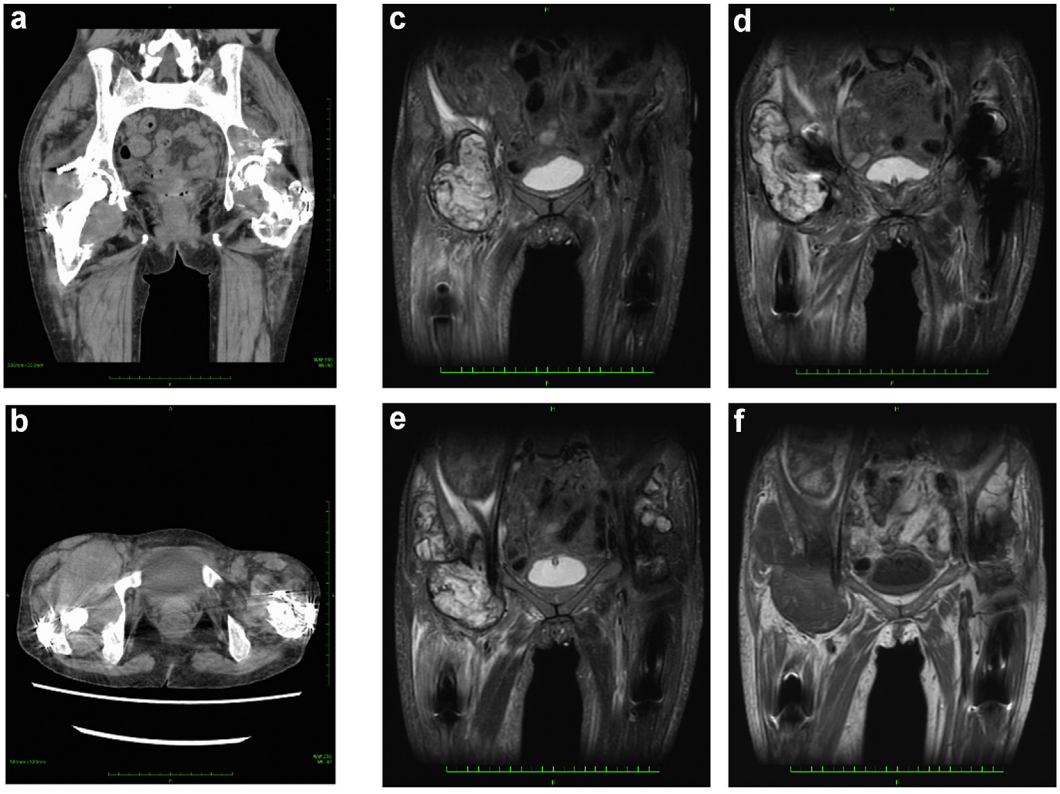

Figure 3. Computed tomography shows bilateral soft tissue masses around the artificial hip joints. (a) Coronal view shows a large mass along the axis of the right thigh and the left ilium. (b) Axial view shows a large mass toward the inguinal region on the right. (c–f) Magnetic resonance imaging performed after 25 years after total hip arthroplasty and development of a mass. (c–e) Presence of low and high signal intensities on T2-weighted coronal images with fat suppression. (f) Isointense or slightly high signals are seen on a T1-weighted coronal image.

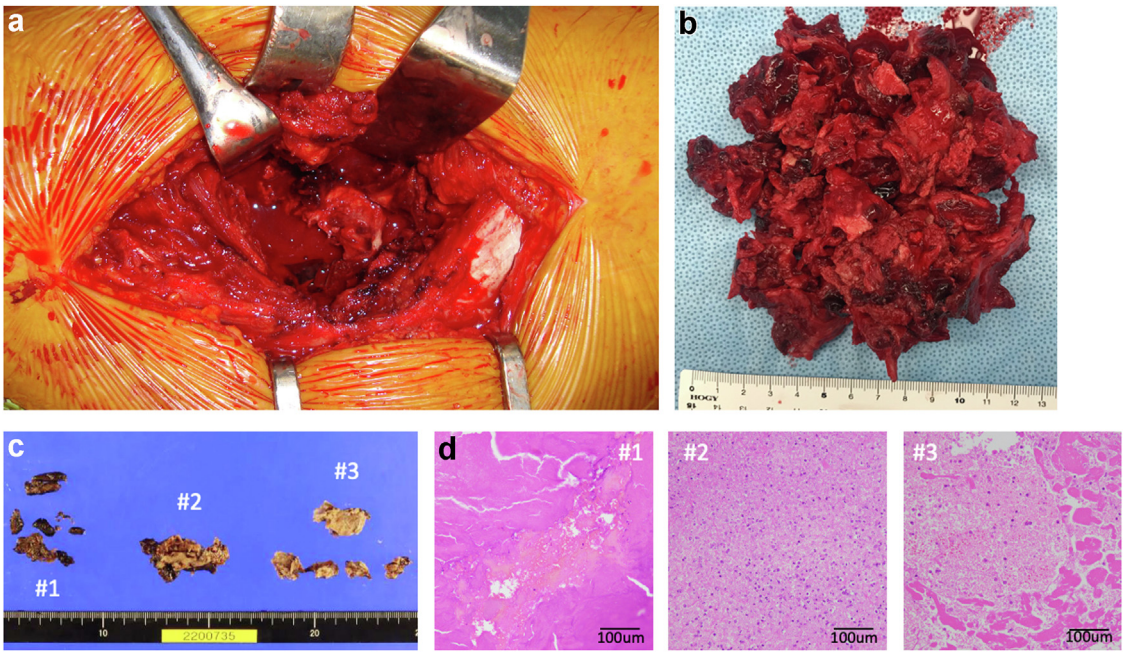

Figure 4. Images showing surgical removal of the mass in the right hip. (a) Surgery was performed with an anterior lateral approach (Hardinge approach). The mass was filled with clotted red blood. (b) The resected mass had a thick fibrous wall surrounding the clots. (c) Macroscopic appearance of the different hematomas observed. (d) Hematoxylin and eosin staining of the resected lesion.

because it is associated with a low risk of recurrence and allows for a reliable histological diagnosis [20-22]. In our case, there was a possibility of infection, and it was necessary to perform the surgery as soon as possible. However, after informing the patient of the potential risk of deterioration in renal function secondary to general anesthesia and the resulting risk of dialysis initiation, the patient refused surgery. Therefore, we decided to perform a minor incision and collect biopsy samples under local anesthesia for both diagnosis and treatment. During the procedure, a large amount of bleeding from the mass was noted. We considered performing an angiographic blood flow evaluation and embolization after dialysis initiation. However, the radiologist determined that the main concern was not arterial bleeding but decreased coagulation function due to renal failure and a potentially poor effect of vascular embolization.

Our case was of advanced osteolysis occurring over a period of 3 years. It was difficult to judge the speed of progression because regular follow-up was not possible and no images were taken during the initial follow-up examinations. If a CEH is found, the orthopedist should ideally follow-up with the patient every 2-3 months and consider resection of the hematoma if the osteolysis progresses. After examining the biopsy specimens, we recommended a hematoma resection for the patient. The patient's pain and symptoms had resolved. However, owing to the burden on daily life due to dialysis initiation and the prognosis of renal amyloidosis, the patient requested a follow-up without further surgical intervention. However, no cases of partial resection of a hematoma have been reported previously, and the evaluation criteria for a postoperative course are unclear. In our case, pain reappeared 6 months after the initial surgery, and the hematoma was resected under general anesthesia. Because only 3 months have passed since then, further follow-up is required to determine whether the CEH will recur. However, while a longer-term follow-up is required to monitor for CEH recurrence, it is also important to assess other long-term outcomes, such as pain, functional status, and quality of life. This case report has not presented data on these outcomes beyond the immediate postoperative period.

Summary

This case report describes a unique and rare occurrence of a CEH after 25 years following THA; this is a significantly longer time interval than that reported previously. This long interval between the initial surgery and the onset of CEH symptoms highlights the importance of considering CEH as a possible cause of anterior hip pain in patients who have undergone THA, even several years after the procedure. Furthermore, the patient in the present case had RA, amyloidosis, and chronic renal failure, which led to an obvious nutritional status imbalance and coagulation disorders. Furthermore, CEH occurs after trauma and invasive procedures. Therefore, considering all of these points, the risk of CEH development in this patient was relatively high when compared with the risk in patients with lesser risk factors. The patient in our case required surgery for extensive osteolysis due to the CEH; therefore, our case report adds to the limited literature on the management of late-onset CEH. By reporting this case, we hope to increase awareness among clinicians of the possibility of late-onset CEH (especially in malnourished patients and in those with potential coagulopathy) and emphasize the importance of prompt diagnosis and management to prevent complications such as osteolysis.

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Conflicts of interest

The authors declare there are no conflicts of interest.

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Informed patient consent

The author(s) confirm that informed consent has been obtained from the involved patient(s) or if appropriate from the parent, guardian, power of attorney of the involved patient(s); and, they have given approval for this information to be published in this article.

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