



Case report

Growth of an intrapelvic pseudotumor associated with a metal-on-metal total hip arthroplasty after revision arthroplasty causing a femoral nerve neuropathy

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ABSTRACT

The development of pseudotumors is not uncommon with metal-on-metal total hip arthroplasty. Pseudotumors that dissect into the retroperitoneal space can cause symptoms of nerve compression. We describe a case of a 53-year-old male with a metal-on-metal total hip arthroplasty who developed mild symptoms of a femoral nerve neuropathy 6 years postoperatively. Revision arthroplasty to a ceramic-on-polyethylene articulation and debridement of the pseudotumor was performed. Postoperatively, the patient's femoral neuropathy progressed and a repeat magnetic resonance imaging showed an increase in size of the pseudotumor despite the removal of the offending metal-on-metal articulation. The patient subsequently underwent a laparoscopic excision of the retroperitoneal pseudotumor. By 17 months post laparoscopic excision of the pseudotumor, the patient's motor deficits resolved, however, sensory deficits persisted in the anteromedial thigh.

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Introduction

The development of pseudotumors is a known complication associated with metal-on-metal total hip arthroplasty (MoM THA). Metallic-wear debris is believed to result in a chronic inflammatory process and a type IV delayed hypersensitivity response from the immune system leading to local tissue necrosis [1]. The prevalence of pseudotumors after MoM THA is estimated to be 0%–6.5% [2]. However, a recent single surgeon magnetic resonance imaging (MRI) review of patients who underwent MoM THA showed that the percentage of patients with detectable pseudotumors could be as high as 68.6% [3]. Pseudotumors often remain subclinical, but they can cause extensive local tissue necrosis often affecting the hip abductors, short external rotators, and flexors. In addition,

there are a number of case reports of large pseudotumors causing compression of intraabdominal structures [4–7].

Compressive neuropathies of the femoral nerve by large pseudotumors have been reported [8–10]. The pseudotumor typically dissects along the iliopsoas bursa into the retroperitoneal space and can result in compression of the femoral neurovascular bundle. Treatment involves revision arthroplasty to address the wear debris generated by the MoM articulation. The cyst can either be excised or decompressed to alleviate the compressive symptoms [4]. To the best of our knowledge, no one has identified cases in which a pseudotumor continues to increase in size even after the removal of the offending MoM arthroplasty. We report the case of a patient with a known pseudotumor and femoral nerve palsy 6 years after undergoing a MoM THA, who subsequently had enlargement of the pseudotumor along with a worsening femoral nerve neuropathy despite a revision to a ceramic-on-polyethylene bearing.

Case history

A 53-year-old Caucasian male presented to us with symptomatic left hip osteoarthritis nonresponsive to conservative management. His medical history was unremarkable and had no known metal

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allergies. His body mass index was 23. The patient underwent a left THA with a cementless Summit stem, cementless Pinnacle acetabular shell, and Ultamet MoM 36-mm bearing (DePuy, Warsaw, IN). The patient's recovery was uneventful.

Six years postoperatively, the patient presented with left leg weakness and anteromedial thigh numbness and paresthesias. His clinical exam revealed decreased sensation in L2 and L3 dermatome distributions along with 4/5 strength in the iliopsoas and quadriceps. Cobalt and chromium levels were elevated at 76.4 and 75.6 ppb, respectively. An AP hip radiograph did not demonstrate signs of component loosening (Fig. 1).

Metal artifact reduction MRI of the left hip revealed 2 heterogeneous lesions. One lesion was isolated within the iliacus and extended into the iliopsoas, measuring $8.8 \times 4.7 \times 9.3$ cm and displaced the femoral neurovascular structures. The second was located posterolateral to the proximal femur measuring $7.8 \times 2.2 \times 5.7$ cm (Fig. 2). Nerve conduction studies revealed no abnormalities; however, electromyography demonstrated decreased recruitment in the vastus medialis and iliopsoas.

In the presence of a large pseudotumor, elevated serum cobalt and chromium levels, and symptoms of a compressive neuropathy, a revision arthroplasty with debridement and head and liner exchange was performed. Upon exposure through a posterior surgical approach, the joint capsule appeared fluctuant and was decompressed, returning 60 milliliters of blood tinged fluid. In addition, there was diffuse synovial staining and the presence of several visible pseudotumors, which were thoroughly debrided (Fig. 3). Intraoperatively, the acetabular cup and femoral stem were well fixed. Examination of the femoral head and taper revealed minimal corrosion and fretting at the head-stem junction. The acetabular shell did not show any signs of wear. The modularity of the Summit stem and Pinnacle Acetabular cup allowed for a head and liner exchange with a BIOLOX delta ceramic femoral head with a titanium sleeve and a moderately crosslinked polyethylene liner



Figure 1. Anteroposterior (AP) hip radiograph performed 6 years postimplantation with metal-on-metal prosthesis in appropriate alignment without signs of loosening, osteolysis, or hardware failure.

(DePuy). Pathology review of the specimen revealed fibrous tissue with marked histiocytic infiltrates and abundant metallic staining of the tissue.

Ten months postrevision arthroplasty, the patient continued to experience progressive numbness and paresthesias in the medial thigh at the level of knee, further instability of the knee with ambulation, and difficulty ascending and descending stairs. A cane and double-upright drop lock knee brace were required for ambulation. Clinically, mild quadriceps atrophy along with decreased sensation in the L2 and L3 dermatomes were present. In addition, a large abdominal mass could now be palpated in the left lower quadrant. Repeat sampling of serum cobalt and chromium ion levels 6 and 10 months post revision revealed a progressive downward trend in ion levels (Fig. 4).

Due to the progression of motor and sensory deficits post-revision arthroplasty and the presence of a palpable abdominal mass, a repeat MRI of the left hip was performed demonstrating a persistent pseudotumor within the iliopsoas and proximal femur. The size of the pseudotumor within the iliopsoas had significantly increased in size, now measuring $9.1 \times 10.2 \times 13.4$ cm, with further displacement of the femoral neurovascular bundle (Fig. 5). In light of the progressive neurological deficits, the decision was made to surgically excise the retroperitoneal pseudotumor.

One-year post revision left THA, the patient underwent a laparoscopic removal of the retroperitoneal pseudotumor. Intraoperatively, the cyst was identified and dissected free from the sigmoid colon. The contents of the cyst were excised and consisted of dark viscous fluid and brown caseous tissue (Fig. 6a and b).

Four months postsurgical excision, symptoms remained unchanged, with quadriceps weakness on ambulation and paresthesias in the anteromedial thigh. Clinical exam revealed mild quadriceps atrophy, and motor strength remained at 4/5 in iliopsoas and 3/5 in quadriceps. Decreased sensation to sharp touch was present in the L3-L4 dermatomes. Nerve conduction studies revealed absent saphenous nerve conduction, and electromyography demonstrated abnormal vastus lateralis and rectus femoris response. Repeat MRI revealed interval resolution of the pseudotumor within the iliopsoas (not shown).

Twelve months after excision of the pseudotumor, quadriceps strength had improved and there was partial return of sensation over the anteromedial thigh. At 17 months post pseudotumor excision, motor deficits resolved and assistive devices for ambulation were discontinued; however, paresthesias in the anteromedial thigh persisted. The patient was able to resume normal functional and recreational activities. At 35 months post pseudotumor excision, the patient continued to have residual paresthesias in the anteromedial thigh.

Discussion

MoM THA have 2- to 3-fold increased rates of revision due to the inflammatory reaction to metal debris leading to the development of macroscopic necrosis, osteolysis, large sterile hip effusions, and pseudotumors [11]. The pathogenesis of this local response to MoM-wear debris has not been fully characterized; however it has been theorized to involve both a cytotoxic and delayed hypersensitivity reaction.

The development of gross instability and need for revision surgery is not the only complications associated with the local tissue response to metallic-wear debris. A number of literature reports describe cases in which pseudotumors associated with MoM arthroplasty cause clinically significant compressive neuropathies involving either the femoral or sciatic nerve [12]. In these instances, prompt revision arthroplasty is advocated [8,9]. The evidence for surgical excision of the pseudotumor is not clear. Some authors

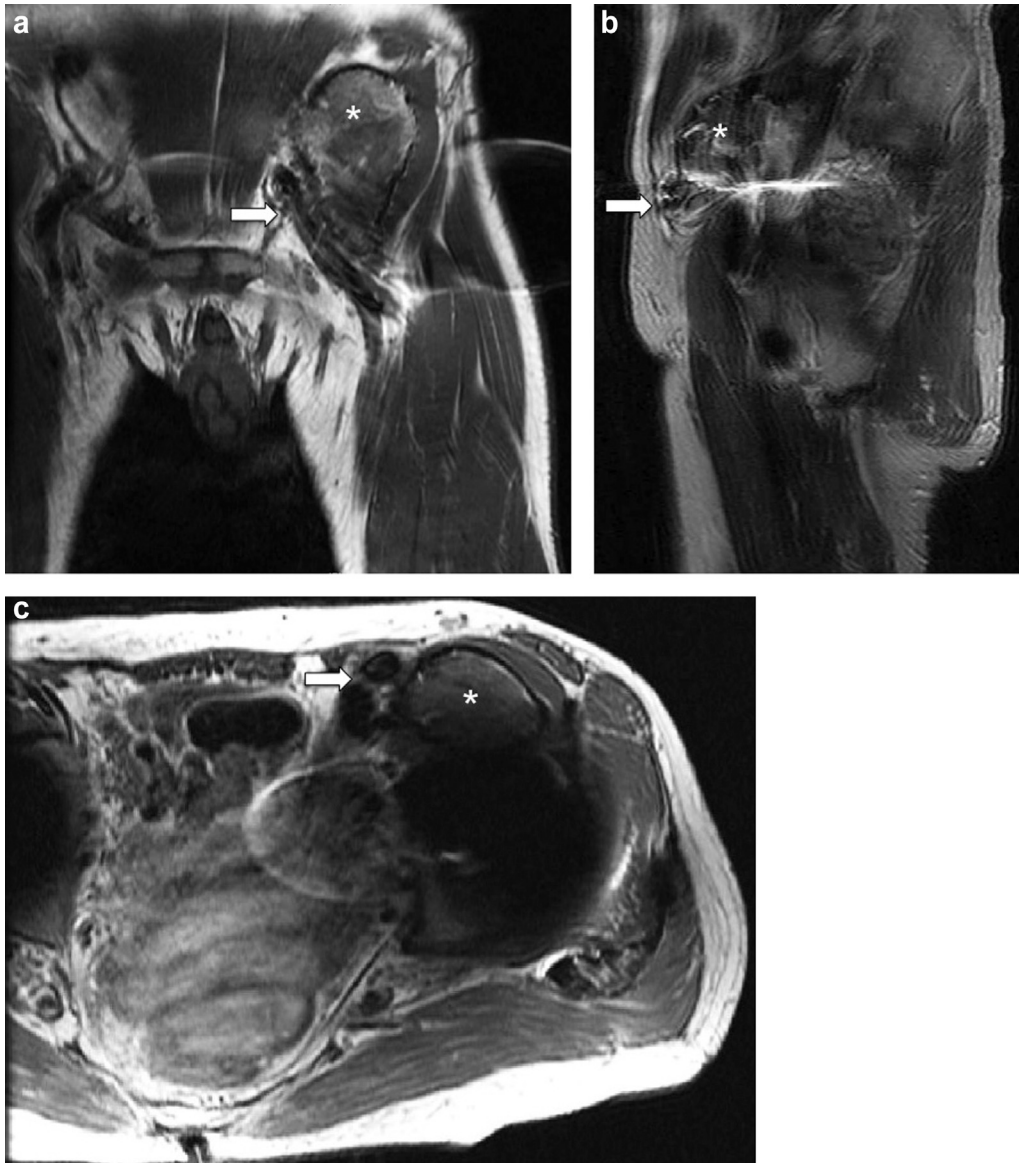


Figure 2. (a) Coronal, (b) sagittal, and (c) axial T1 MRI images of the left hip prior to revision demonstrated a heterogeneous mixed signal lesion within the substance of the left iliopsoas muscle (asterisk). The lesion has a mass effect on the surrounding structures and displaces the external iliac and femoral artery (arrow).

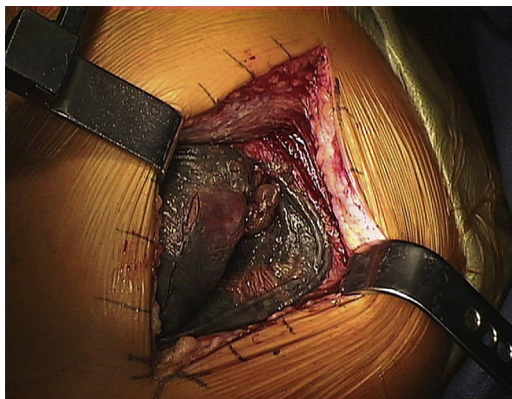


Figure 3. Intraoperative photo of the initial revision arthroplasty with head and liner exchange through a posterior approach to the hip. Upon dissection through the gluteus maximus, diffuse synovial staining was seen in the surrounding tissues.

advocate for complete cyst excision to directly decompress the nerve, which may necessitate either a staged procedure or an extensile surgical approach. Others support debriding as much of the pseudotumor as possible through the same surgical approach as the revision arthroplasty and indirectly decompressing the cyst [12,13]. In our case, the patient presented with a subacute femoral neuropathy with mild motor and sensory symptoms, and thus, we opted to perform a single-staged revision arthroplasty with debridement of the pseudotumor through the same posterior surgical approach. With the removal of the source of metallic-wear debris, it was thought that the pseudotumor would stabilize or regress in size.

We are not aware of any reports of a pseudotumor associated with a MoM articulation continuing to increase in size after the removal of the offending source. In this instance, the pseudotumor continued expand in size and caused a clinically progressive femoral nerve compressive neuropathy with both motor and sensory deficits.

We theorize that the increase in size of the pseudotumor can be in part explained by the cytotoxic effects of cobalt and chromium ions. In high concentrations, both metal ions induced necrosis of

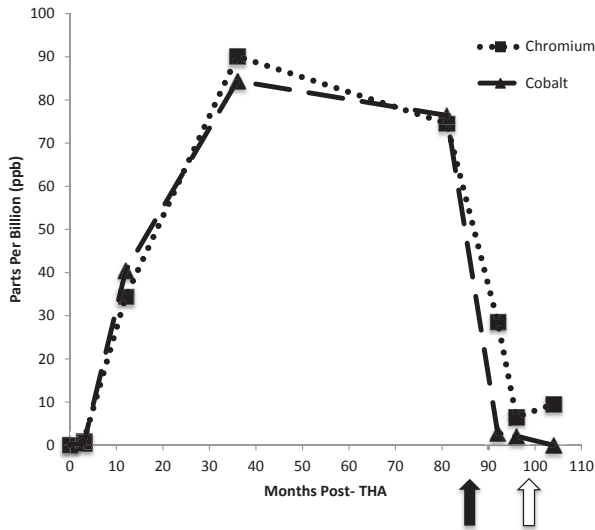


Figure 4. Serum cobalt and chromium levels measured at various intervals following metal-on-metal THA. Revision arthroplasty to ceramic-on-polyethylene bearing was performed 86 months after the primary surgery (black arrow). The retroperitoneal lesion was excised laparoscopically 99 months following the index arthroplasty procedure (white arrow).

macrophages cultured in vitro [14]. The induction of necrosis leads to disruption of the cellular membrane and release of intracellular contents including cytokines, lysosomal enzymes, and phagocytosed metallic-wear debris. The continued presence of inflammatory mediators will again induce macrophage recruitment, phagocytosis of metallic-wear debris, and cell death. This cycle of inflammation within the pseudocyst can lead to continued growth of the pseudotumor [15].

The neuropathy in our patient could have also worsened secondary to a direct effect of metallic-wear debris on the nerve itself. Harvie et al. described 2 cases of a pseudotumor associated with MoM resurfacing arthroplasty causing a dense femoral nerve palsy. In each instance, revision arthroplasty and femoral nerve neurolysis were performed. Intraoperative examination of the femoral nerve revealed a pale flattened appearance that was partially encased within the pseudotumor. Femoral nerve biopsy demonstrated absence of axons and myelin sheaths and large dystrophic calcifications in place of axons [8]. In our case, we did not directly visualize the femoral nerve as it was deeply encased in the pseudotumor making it difficult to grossly examine laparoscopically. Since the patient’s symptoms did improve with removal of the cyst, this suggests that compression of the femoral nerve was likely the resulting cause of the femoral neuropathy; however, other direct effects cannot be discounted.

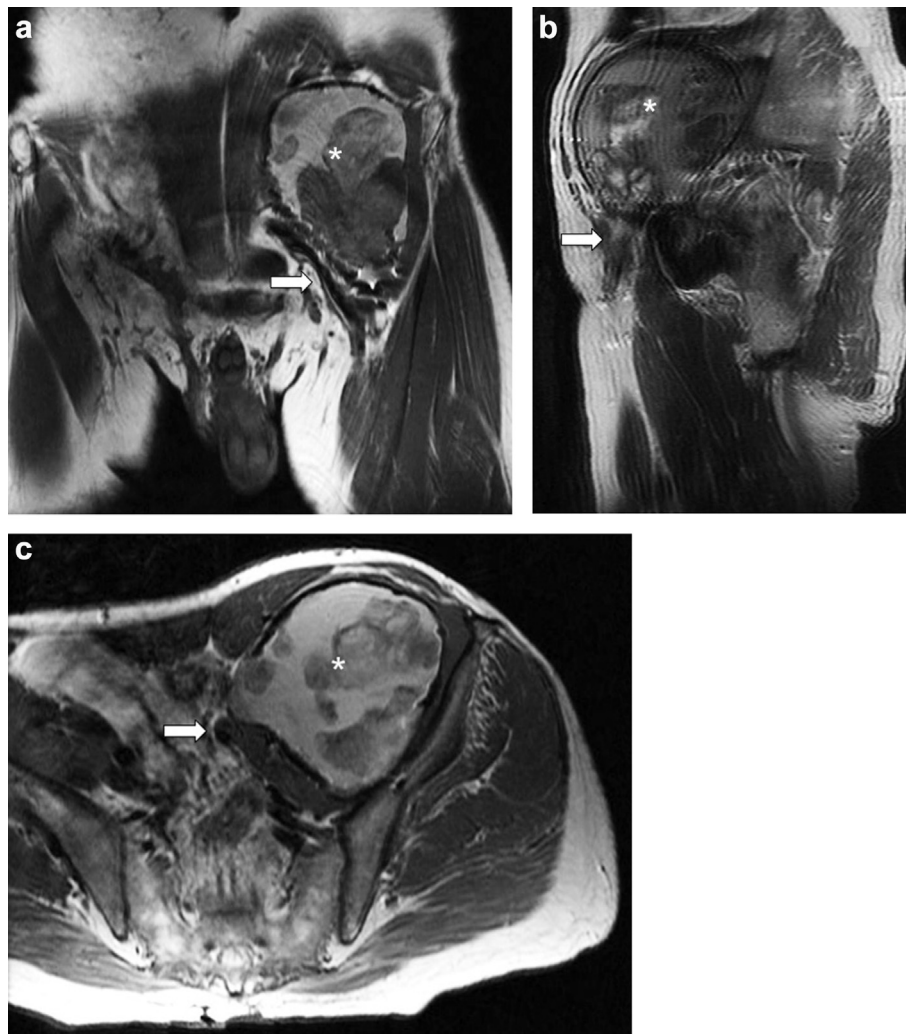


Figure 5. Representative (a) coronal, (b) sagittal, and (c) axial T1 MRI images 1-year post revision revealed a large lesion in the iliopsoas muscle that has increased in size compared to prior MRI (asterisk). Significant displacement of the femoral neurovascular bundle is again demonstrated (arrow). An increase in the fluid component is also noted.

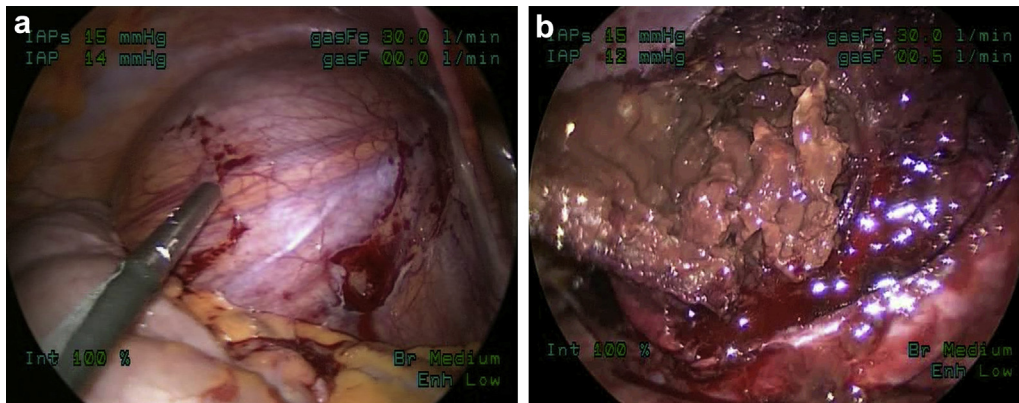


Figure 6. (a) Intraoperative laparoscopic finding of a retroperitoneal pseudotumor which has been dissected free from adjacent sigmoid colon. (b) The pseudotumor consisted of a firm fibrous capsule and contained both cystic areas filled with thick brown fluid and regions of necrotic tissue. All contents of the cyst and the capsule were removed laparoscopically.

Summary

The treatment decision for addressing pseudotumors can be difficult since they have been detected in both symptomatic and asymptomatic patients [3]. Bolognesi et al. proposed an algorithm providing a framework for the evaluation and treatment of patients with MoM THA. This risk stratification algorithm enables orthopaedic surgeons to make objective decisions and reduces the overreliance on just one diagnostic tool [11]. In conjunction with this algorithm, we suggest intrapelvic pseudotumors associated with MoM THA should be closely monitored for symptoms of compressive neuropathies. If symptoms do arise, revision arthroplasty should be considered. There are conflicting reports on whether the pseudotumor needs to be surgically excised in its entirety. The decision on how to address the pseudotumor is case dependent and varies based on factors including degree of nerve compression and ease of accessibility of the pseudotumor. If the pseudotumor is not completely excised one must be cognizant to the possibility that the cyst can continue to grow and cause worsening compressive symptoms. Thus, the presence of a retroperitoneal pseudotumor warrants continued observation even if revision arthroplasty has already been performed.

References

- [1] Willert HG, Buchhorn GH, Fayyazi A, et al. Metal-on-metal bearings and hypersensitivity in patients with artificial hip joints. *J Bone Joint Surg* 2005;87(1):28.
- [2] Wiley KF, Ding K, Stoner JA, Teague DC, Yousuf KM. Incidence of pseudotumor and acute lymphocytic vasculitis associated lesion (ALVAL) reactions in metal-on-metal hip articulations: a meta-analysis. *J Arthroplasty* 2013;28(7):1238.
- [3] Sutphen SA, MacLaughlin LH, Madsen AA, Russell JH, McShane MA. Prevalence of pseudotumor in patients after metal-on-metal hip arthroplasty evaluated with metal ion analysis and MARS-MRI. *J Arthroplasty* 2016;31(1):260.
- [4] Butler RA, Barrack RL. Total hip wear debris presenting as lower extremity swelling. *J Bone Joint Surg Am* 2004;86-A(1):142.
- [5] Hatstrup SJ, Bryan RS, Gaffey TA, Stanhope CR. Pelvic mass causing vesical compression after total hip arthroplasty case report. *Clin Orthop Relat Res* 1988;227:184.
- [6] Madan S, Jowett RL, Goodwin MI. Recurrent intrapelvic cyst complicating metal-on-metal cemented total hip arthroplasty. *Arch Orthop Trauma Surg* 2000;120(9):508.
- [7] Mak KH, Wong TK, Poddar NC. Wear debris from total hip arthroplasty presenting as an intrapelvic mass. *J Arthroplasty* 2001;16(5):674.
- [8] Harvie P, Giele H, Fang C, et al. The treatment of femoral neuropathy due to pseudotumour caused by metal-on-metal resurfacing arthroplasty. *Hip Int* 2007;18(4):313.
- [9] Fokter SK, Repše-Fokter A, Takač I. Case report: femoral neuropathy secondary to total hip arthroplasty wear debris. *Clin Orthop Relat Res* 2009;467(11):3032.
- [10] Schuh A, Werber S, Zeiler G, Craiovan B. Femoral nerve palsy due to excessive granuloma in aseptic cup loosening in cementless total hip arthroplasty. *Zentralblatt für Chirurgie* 2004;129(5):421.
- [11] Bolognesi MP, Ledford CK. Metal-on-metal total hip arthroplasty: patient evaluation and treatment. *J Am Acad Orthop Surg* 2015;23(12):724.
- [12] Fischer SR, Christ DJ, Roehr BA. Sciatic neuropathy secondary to total hip arthroplasty wear debris. *J Arthroplasty* 1999;14(6):771.
- [13] Leigh W, O'Grady P, Lawson EM, et al. Pelvic pseudotumor: an unusual presentation of an extra-articular granuloma in a well-fixed total hip arthroplasty. *J Arthroplasty* 2008;23(6):934.
- [14] Huk OL, Catelas I, Mwale F, et al. Induction of apoptosis and necrosis by metal ions in vitro. *J Arthroplasty* 2004;19(8):84.
- [15] Mahendra G, Pandit H, Kliskey K, et al. Necrotic and inflammatory changes in metal-on-metal resurfacing hip arthroplasties: relation to implant failure and pseudotumor formation. *Acta Orthop* 2009;80(6):653.