Noninflammatory Pseudotumor Simulating Venous Thrombosis After Metal-on-Metal Hip Resurfacing

WERNER MAURER-ERTL, MD; JOERG FRIESENBICHLER, MD; BERNADETTE LIEGL-ATZWANGER, MD; GERDA KUERZL, MD; REINHARD WINDHAGER, MD; ANDREAS LEITHNER, MD

Metal-on-metal hip resurfacing has become a widespread procedure, especially in young, physically active patients. Pseudotumor is a new complication that can occur after hip resurfacing and metal-on-metal total hip arthroplasty (THA).

This article presents a case of a 37-year-old woman who underwent metal-on-metal resurfacing of the left hip for symptomatic osteoarthritis. Twelve months following implantation, the patient reported painless swelling of the left lower leg. There was no clinical evidence of a deep venous thrombosis. Ultrasound and computed tomography showed a solid cystic lesion in the iliopsoas muscle, which communicated with the hip joint and compressed the external iliac vein. As a consequence, the cystic lesion was resected marginally. A few months later, the patient reported some discomfort in the groin and symptoms of instability, metallic clicking, and a restricted range of motion. Clinical and radiological examination revealed normal findings. Determining the serum concentration of cobalt and chromium revealed high increased levels of these metal ions. Ten months following excision of the pseudotumor, the patient reported recurrent swelling of the left lower leg. Computed tomography of the affected area showed a lobulated cystic formation; therefore, a relapse was suspected. At the second revision, the mass was excised and the implant was revised to a conventional ceramic-on-ceramic THA. At 30-month follow-up, the patient was doing well and there were no signs of local recurrence. Additionally, the metal ion levels of cobalt and chromium in the blood had significantly decreased.

Figure 1: Radiograph of the pelvis 6 months after implantation of the ASR Articular Surface Replacement prosthesis, showing the prosthesis in the correct position. Figure 2: Axial CT scan proximal to the hip showing the cystic mass compressing the left external iliac vein (arrow).
Metal-on-metal hip resurfacing has become an accepted and widespread procedure for joint replacement, especially in young, physically active patients. The advantages of this procedure are less bone resection, less stress shielding, more physiological femoral loading, lower risk of dislocation because of the larger head, less leg-length discrepancy, and easier revision. However, the number of revisions for failed resurfacing arthroplasties increased. The most commonly reported reasons for failure are osteonecrosis of the femoral head, fracture of the femoral neck, aseptic loosening, and component malpositioning.

Pseudotumor (estimated incidence at 5-year follow-up: 1%-4%) is a new complication that can occur after hip resurfacing and metal-on-metal total hip arthroplasty (THA). Several reports exist on the development of cystic and solid masses following metal-on-metal hip resurfacing and metal-on-metal and metal-on-polyethylene THA. Pandit et al first described neither malignant nor infective cystic and solid masses associated with metal-on-metal resurfacing and introduced the term pseudotumor. They reported that pseudotumor may be the result of a toxic effect on cells due to particulate wear debris or an idiosyncratic response to the release of metal particles.

This article describes a case of particulate wear debris-laden cystic pseudotumor causing compression of the external iliac vein mimicking deep venous thrombosis (DVT) in a 37-year-old woman who had undergone metal-on-metal resurfacing arthroplasty of the left hip 1 year previously.

**CASE REPORT**

In 2005, a 37-year-old woman underwent a metal-on-metal resurfacing arthroplasty (hybrid fixation) of the left hip for symptomatic osteoarthritis secondary to mild dysplasia. For resurfacing, a porous-coated anatomical system was used (ASR Articular Surface Replacement; DePuy, Warsaw, Indiana). The surgery was performed by a senior orthopedic surgeon using a posterior approach. The patient presented an uneventful postoperative recovery with good functional outcome.

Six months after implantation, the patient reported slight pain in the groin during flexion. Radiographs were normal (cup inclination, 56°; stem shaft angle, 140°; arc of cover, 8.43 mm; leg-length discrepancy, −0.5 cm) (Figure 1).

Another 6 months later, the patient reported painless swelling of the left lower leg. There was no clinical evidence for DVT. Ultrasound and computed tomography (CT) showed a 4.1×2.2×6.0-cm solid cystic lesion in the iliopsoas muscle, which communicated with the hip joint (Figure 2). The tumor mass showed peripheral contrast medium enhancement and compressed the external iliac vein. Phlebography of the left leg also showed compression of the vein. Plain radiographs of the hip showed no signs of implant loosening or dislocation. Consequently, the cystic lesion was resected marginally.

Macroscopy revealed a green–grey intersection side. Inside, the cystic lesion showed distinctive metallosis. Histologically the tissue consisted of fibrous and fibrohyalinized tissue with massive accumulation of macrophages containing visible metal particles (Figures 3A, B). An inflammatory infiltrate or hemosiderin was not seen, and an iron stain was negative (Figure 3C).

A few months later, the patient again reported some discomfort in the groin and symptoms of instability, metallic clicking, and a restricted range of motion.
of motion. Clinical and radiological examination revealed normal findings. Ultrasonography of the iliopsoas muscle showed no local recurrence of the cystic lesion. Determining the serum concentration of cobalt and chromium revealed high increased levels of these metal ions (cobalt, 36 μg/L [standard, 0-0.5 μg/L]; chromium, 30 μg/L [standard, 0-1.9 μg/L]).

Ten months following first revision, the patient reported recurrent swelling of the left lower leg. Computed tomography of the affected area showed a 4.3×2.0×7.7-cm lobulated cystic formation; therefore, a relapse was suspected. At the second revision, the mass was excised and the implant was revised to a conventional ceramic-on-ceramic THA (PinnacleCorail; DePuy). Additionally, a ventrocranial impingement was diagnosed intraoperatively.

At 30-month follow-up, the patient was doing well, and there were no signs of local recurrence. Plain radiographs showed correct position of the implant. Additionally, the metal ion levels of cobalt and chromium in the blood had significantly decreased (cobalt, 0.6 μg/L; chromium, 5.0 μg/L).

**DISCUSSION**

Resurfacing arthroplasty of the hip is technically more demanding than primary THA. Most important is the accurate position of the implant to ensure a low rate of particulate wear debris because resurfacing arthroplasty is less forgiving of malpositioning than THA with metal-on-polyethylene bearings. Additionally, correct implant position prevents prosthesis–bone impingement, soft tissue impingement, or painful local soft tissue reactions due to particulate wear debris. Inflammatory pseudotumors are complications following metal-on-metal hip resurfacing or metal-on-metal THA. Such lesions cause a spectrum of clinical symptoms, ranging from small asymptomatic lesions to massive local infiltrating lesions causing severe symptoms. Large quantities of particulate wear debris lead to soft tissue reactions, which may result in fluid or mass formation. The origins and pathogenesis of such lesions are unknown. Chronic inflammatory reactions in the periprosthetic tissue with pronounced macrophage response, granulomas, perivascular lymphohytic infiltration, high endothelial venules, and extensive soft tissue necrosis following metal-on-metal resurfacing and metal-on-polyethylene articulation have been previously reported. Langton et al reported early failures of metal-on-metal resurfacing arthroplasty due to reactions to metal debris. Davies and Willert et al reported perivascular lymphohytic infiltration of tissue samples from hips with metal-on-metal and metal-on-polyethylene articulation. Davies et al found significant differences in the pattern and type of inflammation seen in periprosthetic tissues, and Willert et al described diffuse, perivascular, and intramural lymphohytic aggregates, which suggest a cell-mediated immune response to particular wear debris (aseptic lymphocyte-dominated vasculitis-associated lesion). In our case, there was a macrophage response to metal debris but no sign of a chronic inflammation or an aseptic lymphocyte-dominated vasculitis-associated lesion.

Park et al reported several cases of early osteolysis following second-generation metal-on-metal resurfacing. Mahendra et al and Kwon et al reported their findings on implant failure and pseudotumor formation. They suggested a delayed-type hypersensitivity to metal (cobalt, chromium, and/or nickel) in the pathogenesis of femoral implant loosening and pseudotumor development. Several cases exist of pseudotumors following metal-on-metal resurfacing or THA. However, such reactions can also be observed following metal-on-polyethylene THA. Despite this, pseudotumors following metal-on-metal THA must be distinguished from those masses.

Glyn-Jones et al posulated that the incidence of pseudotumors tends to increase with time (4% at 8 years). They also found that age (<40 years, P<.001), sex (male<female, P<.001), and implant size (more frequently small-sized implants, P=.008) had a significant influence on the revision rate for pseudotumors. Kwon et al found significantly higher levels of cobalt (P<.001) and chromium (P<.001) in patients with pseudotumors compared to a non-pseudotumor group and a control group. Overall, the woman in our case met all of these criteria.

Clayton et al reported a case of an inflammatory pseudotumor that was associated with femoral nerve palsy following metal-on-metal hip resurfacing. They also suggested an allergic type of hypersensitivity reaction to metal wear debris in the pathogenesis of the newly formed mass. In our case, the patient suffered from bronchial asthma. Therefore, a hypersensitive reaction against metal wear debris could be a plausible explanation for development of the pseudotumor. Nevertheless, a preoperative skin test using platelets made of cobalt and chromium showed no incidence for an allergic reaction against these metals.

**CONCLUSION**

There is no consensus about the origins of pseudotumors following metal-on-metal hip resurfacing. It is likely multifactorial. Further work is needed to understand the etiology of pseudotumors to avoid this serious complication. Additionally, we advise the determination of serum ion levels following metal-on-metal hip resurfacing routinely at clinical follow-up because increment indicates the degree of metal wear debris.

If pain caused by chronic synovitis, metallic clicking associated with instability, or the formation of a pseudotumor is encountered due to metallosis in a patient with metal-on-metal hip replacement, we recommend the change of the prosthesis to a THA with different tribological pairing.

**REFERENCES**


2. Clayton et al.


