CASE REPORT

'Pseudotumour' invading the proximal femur with normal metal ions following metal on metal hip resurfacing

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SUMMARY

A 75-year-old woman who had undergone hybrid metal-on-metal hip resurfacing 8 years earlier underwent revision arthroplasty because of hip, groin and lateral thigh pain. The main differential was aseptic loosening; however, serum cobalt and chromium levels were normal. Multiple imaging modalities revealed a periprosthetic, cystic soft tissue mass adjacent to the proximal femur. A large 'pseudotumour' with proximal femoral invasion was found at revision arthroplasty. We report the first finding of a 'pseudotumour' invading the proximal femur with normal metal ions following metal on metal hip resurfacing.

BACKGROUND

Second generation metal-on-metal (MoM) hip resurfacing arthroplasty has become increasingly popular in young and active patients with osteoarthritis.¹ The procedure preserves the femoral neck and does not invade the femoral canal. The bearing surface consists of a cobalt-chromium-molybdenum alloy (CoCrMo), which has the claimed advantage of lower wear rates when compared to metal-on-polyethylene (MoP).²

However, there have been several reports of an adverse reaction to metal debris resulting in peri-articular masses.³ The mass can be highly destructive locally involving bone, muscle, femoral nerve, sciatic nerve and the bladder.⁴⁻⁶ This soft tissue mass, commonly known as a 'pseudotumour', can cause pain or dislocation leading to revision surgery.^{7,8}

The pseudotumour is believed to be caused by a delayed hypersensitivity response to metal particles.⁹ An association between pseudotumour formation and elevated levels of serum cobalt and chromium has been established.¹⁰

We report a case of a very large pseudotumour invading the proximal femur in a patient with normal cobalt and chromium ions 8 years after MoM hip resurfacing.

CASE PRESENTATION

A 75-year-old woman presented with right-sided hip, groin and lateral thigh pain and the inability to bear weight.

The patient had undergone bilateral hybrid MoM resurfacing (Cormet advanced hip resurfacing system, Corin Medical Ltd, Cirencester, UK) for osteoarthritis in 2005.

In 2009, she presented with left hip pain and underwent revision to total hip replacement

(Taperloc/Exceed/Ceramic Total Hip Replacement, Biomet, Warsaw, Indiana, USA). An intrapelvic pseudotumour was found on MRI and left in situ. The patient had annual follow-up with X-rays and measurement of cobalt and chromium ion levels.

The right hip resurfacing had a 48 mm acetabular component and a 40 mm femoral head component with both articular surfaces consisting of CoCrMo.

The patient presented before her annual review due to sudden increase in pain in her right hip. Two days prior to admission she felt a sudden pain while walking. She attempted to mobilise with two crutches in her home and due to intolerable pain she presented to our department. At the time of presentation she was unable to bear weight on her right hip due to pain.

Examination of the right hip revealed a significantly reduced range of movement limited by severe pain. The range of flexion in the right hip was 0–20° prior to revision. There was no palpable mass in the right groin, buttock or trochanteric region.

INVESTIGATIONS

Plain radiographs showed a well-fixed right hip resurfacing. Below this, starting in the intertrochanteric region and extending distally for 10 cm was a lytic lesion with no significant bone expansion or cortical destruction (figure 1). The white cell count was $7.6 \times 10^9/L$ and C reactive protein was 6 mg/L.

Figure 1 Anteroposterior pelvic view demonstrates hip resurfacing on the right and total hip replacement on the left. There is an area of lucency in the right proximal femoral shaft with attenuation of the bony margins.

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Figure 2 CT axial view showing bilateral periprosthetic soft tissue masses. On the right, the lesion appears adjacent to the anterior right hip joint and also around the lateral part of the femoral neck with partial erosion of the greater trochanter. The left total hip replacement was asymptomatic and therefore not revised.

Serum metal ion levels were within normal range: chromium (Cr^{2+}) 9 nmol/L (normal range 0–10 nmol/L) and cobalt (Co^{2+}) 9 nmol/L (normal range 0–19 nmol/L).

Huge bilateral cystic masses in relation to both hip prostheses were identified on CT (figure 2). The lesion anterior to the right hip joint was seen to breach the posterior cortex of the proximal femur. Metal artefact reduction sequence MRI further characterised the large mixed attenuation lesion lateral to the right greater trochanter of the proximal femur. The masses return mixed complex signal with T2 low-signal thickening of their margins. They were continuous with the lesion in the proximal femur. Within the proximal right femur an expansion of the marrow cavity was replaced by T2 low-signal soft tissue (figure 3A) and T1 intermediate (figure 3B). These findings appeared consistent with osteolysis secondary to metallosis.

Periprosthetic tissues were collected and fixed in 10% buffered formalin and processed into paraffin wax using routine methods. H&E-stained sections were examined using a light microscope. The large mass consisted of necrotic fibrinous material with some chronic inflammatory exudate extending into the skeletal muscle. Sheets of foamy histiocytes were present in the fibrous tissue as well as in some cystic areas, which could have formed as a result of liberation of metal debris from the hip joint.

TREATMENT

Revision was performed via an anterolateral approach to the right hip. There was a large ovoid mass measuring $13 \times 10 \times 6$ cm. It was found overlying the greater trochanter of the femur and penetrated the posterior cortex of the proximal femoral shaft (figure 4). The mass extended posterior to the femur superior to gluteus maximus insertion and anterior to the femoral neck.

A femoral shaft fracture was identified 7 cm distal to the tip of the greater trochanter and a large amount of pseudotumour debris was removed from the femoral shaft. A significant part of gluteus medius was non viable and destroyed by the mass. An intact sciatic nerve was identified adjacent to the posterior aspect of the mass. The mass was completely excised.

An extended trochanteric osteotomy was performed due to the shaft fracture. Following reaming, an uncemented size 8 Arcos modular femoral stem (Biomet, Warsaw, Indiana, USA) was inserted. The proximal femur was reconstructed with a Trochanteric claw (Biomet, Warsaw, Indiana, USA) and Dall-Miles Cable Grip System (Howmedica, Rutherford, New Jersey, USA).

The acetabular component was removed using an Explant (Zimmer, Warsaw, Indiana, USA), which revealed a posterosuperior acetabular defect. Cancellous bone graft was impacted and a 56 mm Regenerex Ringloc cup (Biomet, Warsaw, Indiana, USA) was secured with three cancellous screws. A 36 mm captive polyethylene liner was inserted due to the loss of abductor muscle bulk.

The postoperative course was uncomplicated.

OUTCOME AND FOLLOW-UP

Following revision the patient is seen annually. At her 3-year follow-up the right side was pain free and radiographs were satisfactory (figure 5). She recently had cross-sectional imaging that shows the large left-sided intrapelvic pseudotumour is still present. It causes her intermittent discomfort approximately once every month. The patient has been offered surgical removal of the pseudotumour but at present does not feel her symptoms warrant any further intervention. She will be followed up annually lifelong.

DISCUSSION

During the past 6 years, there have been increasing reports of abnormal soft tissue reactions to MoM hip resurfacings. Pseudotumours, periarticular masses and enlarged bursae have all been reported. The incidence of asymptomatic pseudotumours after MoM resurfacing has been reported to be 8%.¹¹

These pseudotumours are thought to occur due to a reaction to high particle wear and metal hypersensitivity.

Pandit *et al*⁸ described the presence of these periarticular soft tissue masses in a series of 20 metal on metal hips. These locally destructive masses were attributed to an inflammatory response associated with a delayed hypersensitivity reaction to antigen

Figure 3 (A) A T2-weighted MRI coronal view showing osteolysis secondary to metallosis in the right proximal femoral shaft. The soft tissue mass is seen lateral to the greater trochanter. (B) A T1-weighted MRI coronal view showing the soft tissue mass invading the right proximal femur.

Figure 4 Intraoperative photograph demonstrating large 'pseudotumour' found at the right proximal femur.

Figure 5 Anteroposterior radiograph obtained post operatively showing revision of the right total hip prosthesis.

components such as nickel-chromium or chromium-cobalt. These ions can induce apoptosis and necrosis of macrophages as well as lead to a perivascular lymphocyte and plasma cell reaction. This has been described as 'aseptic lymphocyte-dominated vascular associated lesion' (ALVAL).⁹

The periprosthetic tissue obtained in our case demonstrated cystic and necrotic areas. Microscopic evaluation was consistent with this process, showing large areas of histiocytes and fibrin deposition. However, no macroscopic metallosis and no metal wear particles were seen histologically.

Pseudotumours may be asymptomatic and therefore discovered incidentally. The mass can be locally destructive and has been reported to involve the gluteal muscles, femoral and sciatic nerve, bladder and femoral vessels.⁴⁻⁶ Bone involvement is an uncommon finding in the presence of pseudotumours in the context of a MoM hip resurfacing. Hauptfleisch *et al*⁴ performed an MRI study on patients with pseudotumours following MoM hip resurfacing. They found pseudotumours with bony involvement in seven cases. However, they did not report the presence of invasion of the proximal femur into the femoral canal, as described in our case. The linear wear rates at the bearing surfaces of MoM implants is small when compared with MoP implants, however, the number of nano-meter sized particles generated can be up to 500 times higher.² Dissolution of wear particles leads to elevated levels of cobalt (Co^{2+}) and chromium (Cr^{2+}).¹² Modern MoM bearings have also shown uniformly higher serum Co^{2+} and Cr^{2+} levels compared with MoP bearings.¹³ High levels of metal ions are thought to be responsible for adverse reactions such as soft-tissue masses and osteolysis.^{8 14 15}

Bosker *et al*¹⁶ performed a prospective cohort study evaluating pseudotumour formation following MoM total hip replacement. Multivariate analysis revealed that patients with a serum cobalt level $>5 \mu\text{g/L}$ had a fourfold increased risk of developing a pseudotumour. De Smet *et al*¹⁷ analysed serum Co^{2+} and Cr^{2+} levels in 26 patients with MoM hip resurfacing. They found that median serum Co^{2+} and Cr^{2+} levels were approximately 10 times higher in patients with an intraoperative finding of metallosis compared to those without metallosis. The lowest levels of serum metal ions in patients with metallosis were $17 \mu\text{g/L}$ for the Co^{2+} level and $19 \mu\text{g/L}$ for cobalt levels. The serum levels of Co^{2+} and Cr^{2+} in our patient with a very large pseudotumour were substantially lower than the lowest levels of serum metal ions in a patient with metallosis in their study.

This case report highlights several important points. We report the first case of proximal femoral invasion of a pseudotumour following MoM hip resurfacing. The local aggressive nature of pseudotumours should not be underestimated as it can predispose to weakness of the femur and an increased tendency to fracture. We report an important finding of 'normal' cobalt and chromium ion levels with the finding of a very large pseudotumour.

Daniel *et al* demonstrated that serum levels of Co^{2+} and Cr^{2+} peak at 6 and 9 months post-MoM resurfacing. Following this there is a gradual decline in levels due to renal or gastrointestinal excretion.^{18 19} This case demonstrates that the presence of a pseudotumour cannot be excluded on the basis of normal serum cobalt and chromium levels.

Patient's perspective

My pain became worse with time and was resulting in disability. But how could that be when I received the best care and a hip replacement? I was sure that I would not need another hip operation since I was recovering so well. My biggest fear was having another major operation. Then came the clinic appointment, tests and the shock that I had developed a mass spreading into my bone. My first instinct was to wonder whether this was cancer. The surgeons did everything to reassure me that this was likely a benign tumour as a complication of the hip operation nearly a decade ago. I tried to find some more information myself. Yet, there was very little to find online except for professional journal articles. I was perturbed to discover how rare my condition was but after the operation I feel better and was happy to have gone under the knife. I am regaining my independence again, albeit slower than after the first operation. I am just relieved that I do not have to deal with the intractable pain any more.

Learning points

- Pseudotumours are a known complication of metal on metal hip resurfacing arthroplasty, and are thought to occur due to a reaction to high particle wear and metal hypersensitivity.
- Pseudotumours are usually related to high serum concentration of metal ions. In this case, our patient had normal serum cobalt and chromium ion levels.
- Although normally asymptomatic, pseudotumours can cause pain after musculoskeletal infiltration and fractures. This is the first case of infiltration into the proximal femur and femoral canal.

Contributors HK, KS and IA performed the literature review and wrote up the first draft. JS supervised the write up and was the primary surgeon for the patient. All authors approved the final draft.

Competing interests None.

Patient consent Obtained.

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