Pseudotumor in Metal-on-metal Total Hip Arthroplasty: An Unusual Complication

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ABSTRACT
Second-generation metal-on-metal (MoM) total hip arthroplasty (THA) was introduced to overcome the complications associated with the wear debris from ultrahigh molecular weight polyethylene (UHMWPE). However, a new type of complication began appearing, described as aseptic lymphocyte-dominated vasculitis-associated lesion (ALVAL). Patients with ALVAL complain of pain and, in some cases, extensive swelling around the hip joint known as “pseudotumors.” The cause is unknown and probably multifactorial. This may be an adverse reaction to an excess of particulate metal wear debris or a hypersensitivity reaction to a normal amount of metal debris. Surgical findings are typical and symptoms tend to resolve reliably following conversion to an alternative bearing surface. We report one such case of a 55-year-old male, who underwent a revision of bearing surfaces because of ALVAL and pseudotumor formation following an MoM THA at our institute.

Keywords: Aseptic lymphocyte-dominated vasculitis-associated lesion, Hip replacement, Hip resurfacing, Metal-on-metal hip arthroplasty, Pseudotumor, Revision.

INTRODUCTION
Total hip arthroplasty is now an established surgical option to alleviate pain and increase mobility in patients suffering from end-stage primary or secondary arthritis of the hip joint. It is also being done in avascular necrosis (AVN) of the femoral head, which is a more common indication for THA in Indian population as compared with the Caucasian population in whom THA for primary osteoarthritis of hip predominates. According to Frost and Sullivan’s research, almost 70,000 joint replacement surgeries were performed in India in the year 2011. The demand for joint replacement surgery is set to increase further.1

McKee and Watson-Farrar2 introduced the MoM artificial hip joint between 1956 and 1960, and used it in 40 patients with hip disability. They reported 51% good-to-fair clinical results. The system was cementless on both the acetabular and femoral sides. Charnley3 subsequently promoted cemented THA based on the concept of low friction arthroplasty by making use of bone cement for fixation, adoption of a 22.2-mm femoral head, and UHMWPE acetabular cup. Wroblewski et al4 reported mean survival rates of 84.7% at 15 years after surgery in cases where original Charnley-cemented THA was used. However, the outcomes of other cemented THA procedures were very poor: Loosening occurred in 34% at 7.4 years after surgery using the Charnley–Müller type, and 40% at 10 years after surgery using the Müller-type prosthesis.5,6 These poor outcomes of early cemented THAs were due to the implant design and cementing technique in many cases. However, at that time, cement was considered to be the cause and loosening was called “cement disease.”

To circumvent the “cement disease,” there was a shift toward uncemented THA with femoral and acetabular components using MOM bearing surfaces.7 These first-generation MoM prostheses were ultimately taken out of use due to numerous reports of metallosis and patients suffering from a variety of medical conditions due to high levels of metal ions in their blood serum and body tissues. Mechanism of MoM failure was frictional torque leading to subsequent corrosion of the bearing surfaces.8-11 First-generation MoM prostheses were abandoned in favor of bearing surfaces that included polyethylene and ceramic components on the acetabular side. Although ceramic-on-ceramic components have excellent survivorship and have obtained positive results in various studies, it was not without its own problems, such as ceramic fracture, squeaking, and associated osteolysis requiring subsequent revisions in some cases.12,13

Improvements in bearing-surface manufacturing and composition resulted in a second generation of MoM implants with improved wear properties, which
are highly desirable for young and active patients in whom the prosthetics was expected to have greater longevity.\textsuperscript{10,11} It also opened up the opportunity to use larger-sized head in an MoM prosthesis.\textsuperscript{14-17} Early reports suggested high patient satisfaction, low rates of dislocation and wear, and good survival at medium-term follow-up.\textsuperscript{18} Subsequently reports of the formation of periartricular masses in some patients, referred to as pseudotumors, started appearing. It is also described as ALVAL. The term is currently used to denote a periartricular mass caused by an immunological delayed hypersensitivity response to metal particles, characterized by a lymphocyte-dominated histological pattern.\textsuperscript{19} A more specific and broader term used is adverse reaction to metal debris.\textsuperscript{20} The clinical picture may vary from an unexplained discomfort in the hip region, sometimes accompanied by clicking or subluxation.\textsuperscript{21} Occasionally on examination, a swelling in the buttock region or proximal thigh or a lump in the inguinal region can be found. Imaging studies, such as magnetic resonance imaging (MRI) help in making a diagnosis and planning of surgery. When the diagnosis of pseudotumor is established and symptoms are troublesome,\textsuperscript{22} revision surgery is an option. The mass is removed as radically as safely possible and the bearing surfaces are revised.

\section*{CASE REPORT}

In January 2017, a 55-year-old man was admitted to our institute with complaints of occasional left-sided posterolateral thigh and hip discomfort with swelling in his gluteal and trochanteric region for the past 3 to 4 months. The swelling was ill-defined and cystic in nature with no signs of inflammation. He had undergone successful uncemented THA in May 2009 for AVN of the left femoral head in which large-diameter femoral head MoM prosthesis was used. The acetabular cup and metal insert were of 56 mm size and femoral head of 36 mm (DePuy Synthes PINNACLE\textsuperscript{®} Hip Solutions). The contralateral hip also with AVN of the femoral head was replaced with a similar prosthesis after 2 months of the first replacement and since then, he has remained symptom-free until date.

Fresh radiographs of the affected hip and pelvis with both hips were carried out that revealed osteolysis in inferior aspect of acetabulum, ischium, and proximal femoral and trochanteric region (Fig. 1).

A retrospective analysis of his radiographs revealed that until 6 years of his follow-up, the prosthesis appeared secure with no signs of loosening or zones of osteolysis (Fig. 2).

As there was no history of fever or local signs of inflammation apart from swelling to suggest infection, a provisional diagnosis of ALVAL and pseudotumor was made, based on its not-so-rare occurrence in previous studies in literature on MoM THAs.\textsuperscript{23}

Further imaging investigations were done in order to confirm our provisional diagnosis. The MRI of the pelvis with both hip joints (Figs 3 and 4) demonstrated a soft tissue lesion in the left periarticular region extending into thigh posteriorly. The size of the lesion was reported as 95 × 57 mm with edema in the posteromedial regions of the hip and also in the gluteal muscles. The likely differential was provided as pseudotumor after clinical correlation. Routine blood investigations including cell counts, erythrocyte sedimentation rate, and quantitative C-reactive protein were normal.

Patient was planned for revision of the bearing surfaces according to the recent guidelines for patient management with MoM arthroplasty-related adverse reactions.\textsuperscript{24} In the operating room, after induction of general anesthesia, the patient was placed in lateral position and the previous incision of the posterolateral

\begin{figure}[h]
\centering
\includegraphics[width=\textwidth]{fig1.png}
\caption{X-ray showing areas of lysis (red arrows) in the inferior aspect of acetabulum and in the proximal femoral and trochanteric region.}
\end{figure}

\begin{figure}[h]
\centering
\includegraphics[width=\textwidth]{fig2.png}
\caption{X-ray of pelvis with both hips showing well-fixed implants (left side) at 6 years follow-up.}
\end{figure}
exposure to hip joint was utilized. The abductors were found to be normal and partial engulfment of the short external rotators by the mass was seen. The abnormal mass-like tissue was excised as much as possible (Figs 5 and 6) and sent for complete histopathological evaluation. During the revision procedure, the patient’s acetabular and femoral components were found to be well fixed. No evidence of corrosion was noted in the femoral head and at the neck taper of the femoral component. The revision was to a ceramic-on-polyethylene articulation with a highly cross-linked polyethylene acetabular liner and a ceramic femoral head. The insert used was of standard 56 mm size and femoral head of 36 + 8.5 mm (DePuy Synthes) size was found suitable.

Postoperatively, patient was made to stand and walk with help of a walker on the next day of the surgery. Appropriate physiotherapy exercises were initiated and monitored. Hip precautions were explained. The patient was relieved of the groin and thigh pain, which he had felt earlier before undergoing the revision surgery. Patient was discharged at stitch removal on 12th day after surgery.

The histopathological report confirmed our provisional diagnosis. The microscopic findings (Fig. 7) showed multiple soft tissue pieces lined by hyperplastic synovial lining, largely covered with necrotic fibrinous exudates. The underlying fibrous tissue showed presence of sheets of macrophages along with lymphocytes and congested blood vessels along with few histiocytic giant cells. The final impression was consistent with a diagnosis of ALVAL.

**DISCUSSION**

Second-generation MoM prosthesis was introduced for its advantages over conventional articulations, such as low rates of wear and increased stability due to larger head size. The use of this prosthesis had become increasingly popular during the last decade, especially...
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in young, mobile patients. However, there have been several reports of high incidence of ALVAL and pseudotumor formation in patients who underwent MoM THAs and, more recently, in patients undergoing MoM hip resurfacings.

The possible mechanism behind ALVAL in MoM THAs is the slow release of metal ions from the prosthetic-bearing surfaces as a by-product of normal wear. These metal ions are present in adjacent peri-prosthetic soft tissue. These wear particles elicit a type IV response in the local tissue. This local destructive response can lead to pain, osteolysis, loosening of the prosthetic components, and formation of soft tissue mass. Though overall wear rate and size of the wear particles from the second-generation implants are smaller than those associated with the first-generation MoM prostheses or from conventional metal-on-polyethylene components, it is postulated that the smaller size and higher surface area of MoM debris particles actually facilitate their diffusion into surrounding tissues and local tissue reaction resulting in the formation of pseudotumors.

Symptoms emerge slowly and, initially, are mild and often have little impact on patient’s activities. When symptoms of swelling, pain in the groin, and clicking become sufficient to warrant revision, a sizeable pseudotumor with extensive local-tissue destruction may already be present. A high index of suspicion of ALVAL is also warranted in young, mobile patients. However, there have been several reports of high incidence of ALVAL and pseudotumor formation in patients who underwent MoM THAs and, more recently, in patients undergoing MoM hip resurfacings.

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Symptoms emerge slowly and, initially, are mild and often have little impact on patient’s activities. When symptoms of swelling, pain in the groin, and clicking become sufficient to warrant revision, a sizeable pseudotumor with extensive local-tissue destruction may already be present. A high index of suspicion of ALVAL is also warranted in cases presenting with symptoms even after a very long time of the primary surgery of MoM THA. In our case, patient developed symptoms almost 7 years following his replacement surgery. A search of literature reveals that pseudotumors have been reported to become symptomatic up to 15 years postoperatively. More serious symptoms include dislocation, nerve palsy, and fracture, depending on the location and size of the pseudotumor.

Due to these possible complications, regular follow-up and a high index of suspicion for a metal-related adverse reaction are very important. Imaging modalities like ultrasound and computed tomography scan provide some information, but it is the metal artifact reduction scanning MRI that helps in confirming a diagnosis. The ALVAL is confirmed by histopathology of the biopsy specimen.

Since a large number of patients, especially in the younger age group, underwent hip arthroplasty with MoM-bearing surfaces, one can anticipate an increased number of patients presenting with ALVAL and undergoing subsequent revisions for prosthetic failure. Accurate assessment is crucial because an intraoperative diagnosis of chronic inflammation suggestive of ALVAL will necessitate a replacement of the prosthetic component-bearing surfaces. Also, the importance of intervening early cannot be understated. Late intervention in such cases when gross osteolysis and loosening has already occurred with an unstable prosthesis and inadequate bone stock can lead to a difficult revision surgery.

REFERENCES


