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CASE REPORT

Massive Pseudotumor in a 28mm Ceramic-Polyethylene Revision THA: A Case Report

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Abstract

This report reviews the findings of a massive pseudotumor detected pre-operatively in a 13-year-old revision total hip arthroplasty. The case is unique in that the bearing involved was a 28mm zirconia ceramic head on a polyethylene liner. We propose that the pseudotumor arose from ultrafine titanium particles liberated from the proximal porous coating of the femoral stem. We suspect that the osteolysis produced from polyethylene wear exposed the proximal porous coating and, via a process of mechanical abrasion with the surrounding soft tissues, liberated ultrafine titanium particles. We believe the pseudotumor formed because the patient was pre-sensitized to metal debris based upon a pre-operative lymphocyte T-cell proliferation test (LTT). Based upon this unique case, we feel that pseudotumors more likely form when there is a high rate of ultrafine metal particles generated in a pre-sensitized patient. Finally, we introduce what we believe are the main biologic wear responses in THA. Further research is needed to validate this proposed model.

Keywords: *pseudotumor, ceramic, polyethylene, osteolysis, THA, bearing wear eesponse, titanium debris*
Level of Evidence: *AAOS Therapeutic Study Level IV*

Introduction

Over the last decade, pseudotumor has become a rising complication of total hip arthroplasty (THA). It was first described by Griffiths in 1987 in a series of 15 patients with metal-on-polyethylene THA. [8] The presentation of pseudotumor varies, ranging from asymptomatic cases accidentally observed during routine follow-up, to a patient with well-fixed implants having pain, to severe osteolysis with implant failure requiring complex revision arthroplasty. Although the pseudotumor response was first described in metal-polyethylene implants, the more recent literature of the last 10 years impugns metal debris as the pro-inflammatory nidus for pseudotumor formation. [1,5,6,7]

We report a case of a massive pseudotumor that arose in a revision THA with a ceramic-polyethylene bearing. Based on intra-operative observation and review of histologic tissue, we propose a mechanism of pseudotumor formation in this case.

Case Review

HISTORY

This case involves a 59-year-old female suffering from avascular necrosis of the hips. She has idiopathic thrombocytopenia and has no other risk factors for avascular necrosis. She had a spleenectomy at age 27. For the right hip, she had a core decompression at age 30. Her right hip condition still remains stable with only mild intermittent pain.

The left hip was treated with primary THA at age 30 (May 1984). Her reconstruction provided a good functional lifestyle, allowing her to enjoy life as a mother. At 15 years post-op she began having pain and a limp. She

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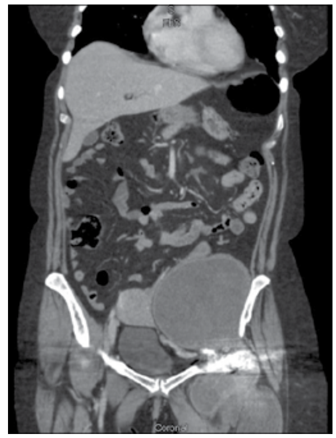
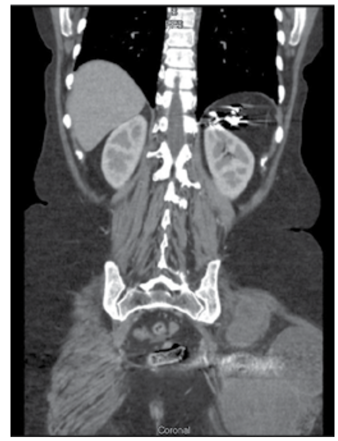
underwent revision THA in June 2000 (16 years post-op) for osteolysis and mechanical loosening of her implants. On the acetabular side, the patient was revised with a porous plasma spray modular titanium cup (Vision™ , Biomet, Warsaw, IN) with 3 screws. The acetabular liner was a compression-molded polyethylene cup (Himont 1900 UHMWPE) with a 10 degree posterior hood placed inferiorly and posteriorly. Her stem was revised with a titanium alloy modular revision stem (Modular Reach™ , Biomet, Warsaw, IN). The head was a 28mm zirconia ceramic bearing (CeramTec, Plochingen, Germany). Post-operatively, she recovered with no problems and again enjoyed an active lifestyle as a mother of 3 children.

She suffered one late dislocation in 2007 which was treated with a closed reduction in the emergency room. She had no subsequent dislocations.

At 9 years post-op from her revision THA, the patient noted no pain or problems with her hip on annual review. The patient was then seen at 10.5 years post-op. She reported suffering from mechanical low back pain and mild left hip pain. At the time she started a weight loss program with daily exercise and had lost 30 pounds. Eccentric polyethylene wear was noted radiographically. Her hip exam showed no hip irritability. She was started on a lumbar trunk stabilization program and her hip was observed. The patient returned at 12 years post-op and was symptomatic in her left hip. She reported hip clicking with flexion and had activity-related pain and mild hip “fullness.” Her hip range testing was comfortable with passive range, but her mid-thigh circumference at that time was 2cm greater on the left. Radiographs showed increased eccentric polyethylene wear. At that time a modular bearing exchange and debridement surgery of the hip was recommended. No other radiographic studies were ordered. The patient declined surgery to finish her teaching duties at an elementary school for the upcoming year.

At 13 years post-op the patient returned with increased pain and swelling (Figures 1a-1b). She had developed numbness and tingling in her left leg. An MRI of the lumbosacral and upper pelvis was performed to evaluate for sciatica. Two tumors were seen on this study which prompted further studies by her local physician. These included MRI’s of the pelvis and thigh and a CT scan of the pelvis and chest. Loculated masses were identified within the pelvis, hip, and thigh; all appeared to emanate from the hip region (Figures 2a-2c). In addition, one mass was seen extending to the anterolateral distal thigh. Her hip exam revealed only mild irritability. Her mid-thigh circumference was now 6cm greater on the left.


Figures 1a-1b*. Preoperative radiographs of revision left THA. These are taken 13 years post-operative from the patient’s revision THA.*
Figure 1a*. AP Radiograph. Note the significant swelling medial to the hip and superior to the greater trochanter. Notice the osteolytic bone loss of the medial femoral neck region.*
Figure 1b*. Lateral Radiograph. This image demonstrates the significant periarticular soft tissue swelling and peritrochanteric bone loss.*


Figures 2a-2c*. Pre-operative coronal CT scan demonstrating extent of pseudotumor emanating from left THA.*
Figure 2a*. Coronal CT cut posterior to the left hip joint. Notice the large pseudotumor extending within the gluteus maximus back towards the iliac crest.*
Figure 2b. Coronal CT cut at the level of the left hip joint. Note the extent of the pseudotumors within the pelvis, under the gluteus medius, and the lateral region of the gluteus maximus.
Figure 2c*. Coronal CT cut anterior to the left hip joint. Note the enormity of the pseudotumor within the pelvis. Also note the anterior pseudotumor which tracked along the medial femur.*

A hip aspiration was performed, drawing off 175cc of thick, dark fluid with a dark brown and maroon coloring. There was a normal string sign. All cultures were negative. These included aerobic, anaerobic, fungal, and mycobacterium cultures. Fluid analysis showed a red cell count of 840,000 and a white cell count of 1,000 with 58% neutrophils, 32% lymphocytes, and 10% monocytes. Serum C-reactive protein was mildly elevated at 1.6mg/dL (normal <0.3) and erythrocyte sedimentation rate was 32mm/hr (normal 0-15). Her CBC was normal. Serum blood was drawn for a metal lymphocyte T-cell proliferation test (LTT) which was sent to Orthopaedic Analysis. [10,11,19,39] Results showed moderate sensitivity to nickel metal particles (Figure 3).

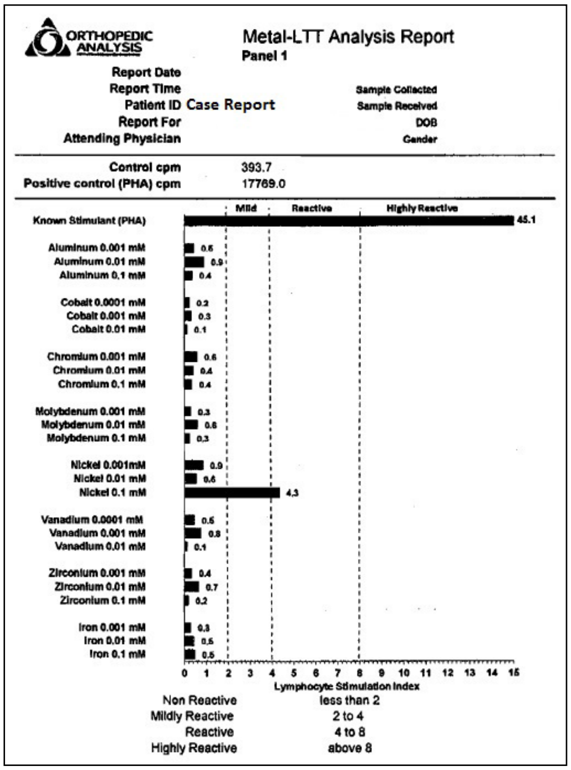


***Figure 3**. Graphic display of Lymphocyte T-cell Proliferation Test (LTT) for metal sensitivity. This patient showed moderate reactivity to Nickel particles at 0.1mM concentration.*

INTRA-OPERATIVE FINDINGS

Intra-operative examination showed a large multiloculated mass that was extending in multiple directions surrounding the hip region. A large 8x14cm mass was lateral to the greater trochanter and extended superiorly within the gluteus maximus. Upon entering the mass, the fluid exited under considerable pressure, shooting out approximately 20cm in distance. The fluid was bloody with a coloration of dark brown and maroon (Figure 4).

***Figure 4**. Intra-operative photograph of left hip pseudotumor upon opening the iliotibial band and splitting the gluteus maximus muscle. Photograph is of the patient in lateral decubitus position with the head to the right of the photo. Note the bloody fibrinous material within the cyst.*

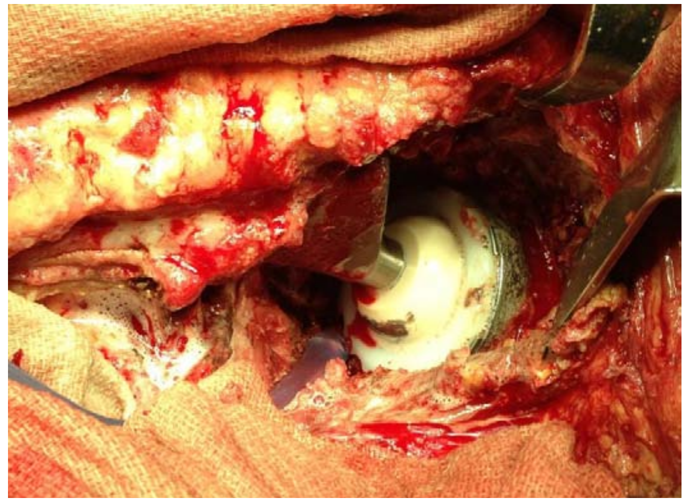


***Figure 5**. Intra-operative photograph of left THA in-vivo after excision of posterior and lateral pseudotumors. Significant osteolytic bone loss is seen around acetabulum and proximal femoral stem. Also note the metal smear on the zirconia ceramic head located inferiorly. This was caused by the patient's single hip dislocation 6 years prior. Despite significant osteolysis, both implants were solidly fixed to bone.*

The hip showed implants that were well affixed to bone with porous coating (Figure 5). Severe osteolytic bone loss was noted in the proximal femur down to the metadiaphyseal region. The cup showed several osteolytic holes.

Five major pseudotumor masses were seen. One pseudotumor mass extended along the iliopsoas into the pelvis for a distance of 12cm (Figures 6a-6b). Another mass extended along the femur and under the vastus lateralis to the distal one-third of the thigh. The third mass extended in between the lateral ilium and gluteus medius up to the iliac crest. The lateral peritrochanteric mass extended posteriorly over the gluteus maximus. Finally, the fifth large mass extended down the medial adductor for a distance of 7cm.

The pseudotumors were excised and a modular bear-

***Figures 6a-6b**. Intra-operative photographs showing delivery of pelvic pseudotumor into the hip. Photographs show the femur reflected anterior to the acetabulum. Views are of the left hip in the lateral decubitus position.*

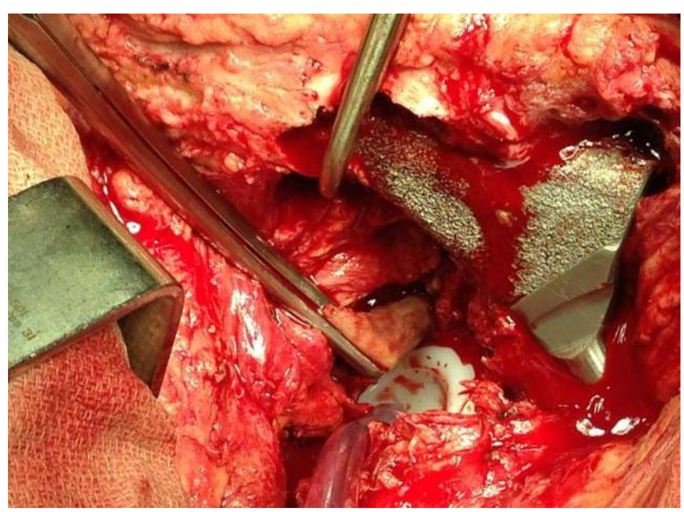


***Figure 6a**. Intra-operative photograph. An aortic cross-clamp is seen curving into the pelvis along the iliopsoas. The clamp is grasping the inner wall of the intra-pelvic pseudotumor pulling it inferiorly.*

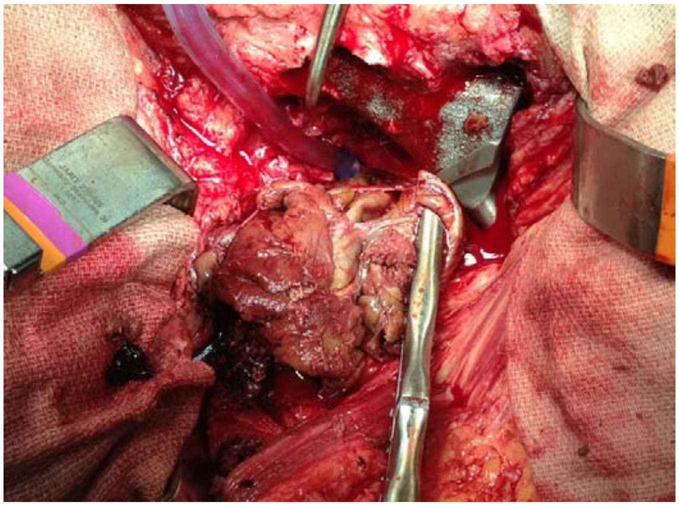


Figure 6b. *Intra-operative photograph. With the assistance of a long rongeur and the aortic cross-clamp, the intra-pelvic pseudotumor is dissected off the iliopsoas with Metzenbaum scissors and delivered into the hip region.*

ing exchange was performed (Figure 7). The femoral taper showed no corrosion or adverse wear (Figure 8). A highly cross-linked, vitamin E infused polyethylene cup (Biomet, Inc., Warsaw, IN) was used. The head was changed over to a 36mm Delta ceramic head with an internal titanium sleeve (CeramTec, Plochingen, Germany). At 1 year post-op the patient still has a mild gluteus medius lurch, but remains pain free.

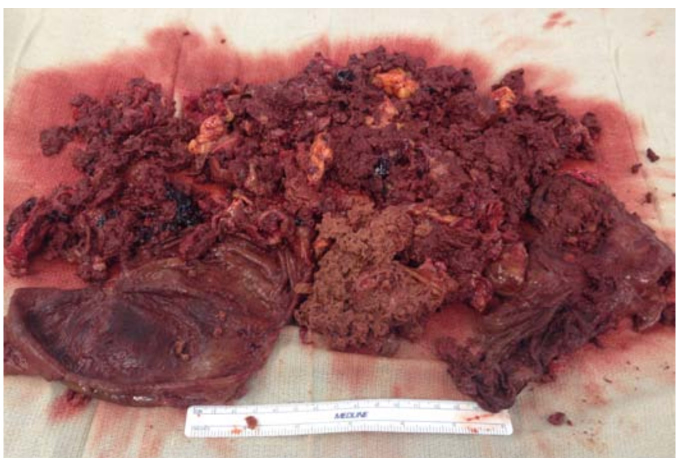


Figure 7. *Photograph of gross specimen recovery of all pseudotumors from the left pelvis, hip, and thigh. Ruler placed inferiorly is 15cm in length.*

Figure 8. *Intra-operative photograph of modular taper junction of the femoral stem. The ceramic head did not have an internal metal jacket. The taper junction shows no abnormal abrasion and is free of corrosion.*

HISTOLOGY

The histologic examination of the pseudotumor was obtained near the base of the intragluteal pseudotumor, just superior to the acetabulum. It was a representative sample of all five pseudotumor masses resected. The histologic images are presented in figures 9a-9b. Within the cyst there contained old, decaying red blood cells and fibrin clots. The wall of the pseudotumor was thin, measuring 1.5 to

Figures 9a-9b. *Photomicrographs of histologic specimens of pseudotumor. Specimens were preserved in formalin and processed with Hematoxylin and Eosin (H&E).*

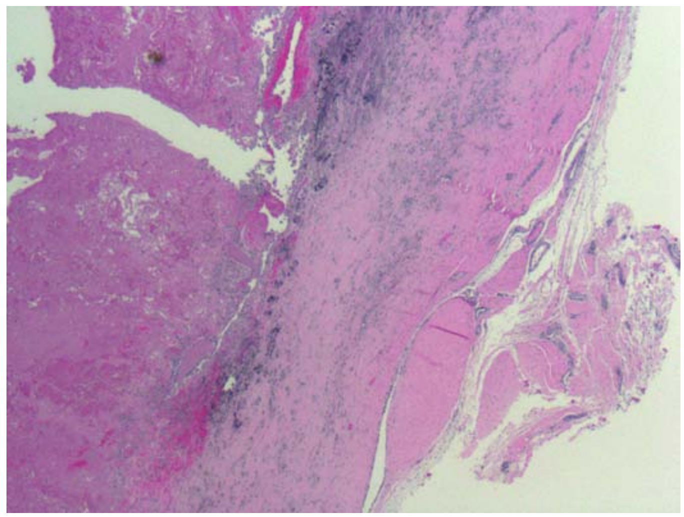


Figure 9a. *4x magnification of pseudotumor showing that the wall itself measures 1.5 to 3mm. Within the cyst there is decaying RBC's along with fibrin clots (left side). One can see the collagen matrix and fibroblasts that form the pseudotumor sac. The inner lining consists predominantly of monocytic histiocytes. There is no lymphocytic response seen within the pseudotumor or in the perivascular regions.*

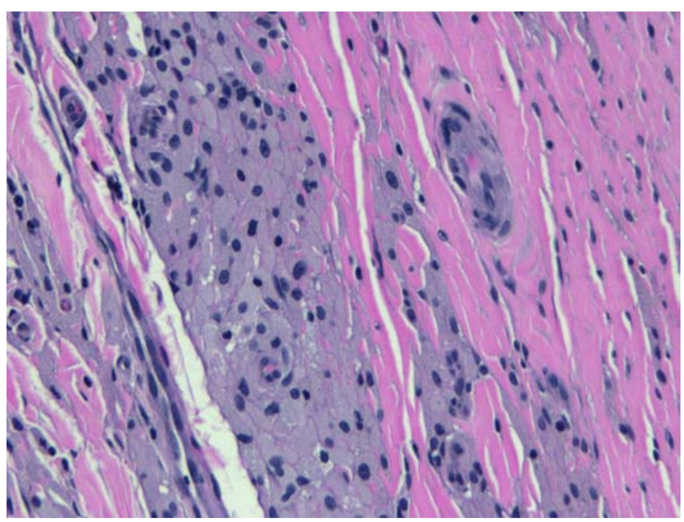


Figure 9b. *40x magnification of pseudotumor wall near its inner surface. Notice specifically the ultrafine light grey/bluish colored particles within the histiocytes. The particles do not refract, indicating that the particles are not polyethylene debris. Instead, these ultrafine particles are likely a titanium alloy particulate shed from the proximal porous coating of the femur.*

3mm, and consisted primarily of collagen fibers aligned haphazardly and interspersed with fibroblasts. The inner lining of the pseudotumor predominantly contained monocytic histiocytes. There was a paucity of giant cells. Furthermore, there was no lymphocytic response within the pseudotumor wall or perivascular vessels. [3,9,10,37] The histiocytes contained ultrafine titanium metal particles (there was no other metal alloy in the hip construct). [29] Some histiocytes contained hemosiderin pigments likely acquired from the decaying RBC's within the cyst. No particles were seen freely interspersed within the collagen matrix. Vascularity to the pseudotumor wall came from local connections from the outer wall to the surrounding muscle. [32,33]

Discussion

This case of pseudotumor formation is unique. The senior author (ejm) has an extensive history of treating metal-metal bearing associated pseudotumors and this case was by far the most extensive pseudotumor he has treated. In all, five large pseudotumor masses were excised in a 5-hour long operation. The bearing in this case was a small-diameter head (28mm) made of zirconia ceramic. The eccentric wear was visually evident, but could not be described as excessive. In addition, we carefully examined the head-taper junction and did not visually observe any taper corrosion reaction nor adverse metal wear. [13] We feel in this case that the osteolysis produced from polyethylene wear exposed the proximal porous coating and, via a process of mechanical abrasion with the surrounding soft tissues, liberated ultrafine titanium particles. Histologically, the histiocytes in the pseudotumor contained ultrafine metal particles. Since no other metal was used in the case, we must conclude that the metal debris derived from the titanium implants.

Most surgeons currently believe the metal debris causing pseudotumors derives from cobalt-chrome alloy bearings. [14,15,16,18,19] This is based upon the wear debris phenomenon seen with metal-metal bearings over the last decade. [20,22,24,27,28] The toxic reactive synovitis seen with this bearing can cause effusion, pain, and, in some cases, pseudotumor formation when the bearing couple is improperly designed or mated.

A pseudotumor reaction consists of an expanding extra-capsular inflammatory process consisting of collagen, fibroblasts, and, in this case, histiocytes. Anecdotal evidence provided by older arthroplasty surgeons described this phenomenon, verbally, as far back as 27 years ago and was associated with metal-polyethylene hip bearings. [8,35] Since polyethylene induced osteolysis is well described and consistent in presentation, the early and later descriptions suggest metal debris as the initiator of pseudotumor formation. The pseudotumor reaction, therefore, may reflect the interaction of an overactive immune system in combination with metallic particulate debris. [10,39]

In this case we call into question the effect of porous coating as a contributor to the particulate metallic load within the effective joint space. Many porous coatings on titanium alloy stems are known to shed debris. [20,38] Furthermore, exposed porous coated surfaces resulting from classic PE-induced osteolysis can release increasing amounts of metal as the area of exposed porous coating increases. In this case the sequence of pseudotumor formation followed an escalating course of periprosthetic osteolysis. We propose that the sequence of events forming the pseudotumor syndrome is the following: (1)PE-induced boney osteolysis eroded the proximal femoral metadiaphysis [4,17,23,25]; (2)the exposed prosthetic porous coated surface mechanically abraded with the soft tissues introducing ultrafine particulate debris into the effective joint space [21,26,30,36]; (3)the patient's pre-sensitization to metal (positive LTT results) triggered inflammatory cytokines to form the pseudotumor capsule. [19,28,31,34]

This case illustrates the point that a pseudotumor can form in the absence of cobalt-chrome implants. A majority of reports in the last decade have impugned cobalt and chrome metal particles of eliciting an immune response unique to that metallic alloy. [10] Based upon this case, we reject that concept outright. We believe that the biological response to particulate hip debris can be categorized into five main syndromes (Figure 10). These biologic responses are based upon particulate type and size, rate of particle formation, and pre-sensitization of the internal immune system to metal debris. [2,12,13,20]

In retrospect, to mitigate the recurrence of the pseudotumor, we advocate sealing off the exposed porous coating to minimize metal particle generation. We feel this would be best accomplished by covering the exposed porous coating with methyl methacrylate cement. In the future, based on the findings in this case, we intend to cover all exposed porous surfaces or, if possible, to replace modular porous segments with segments which have smooth surfaces (preferably polished).

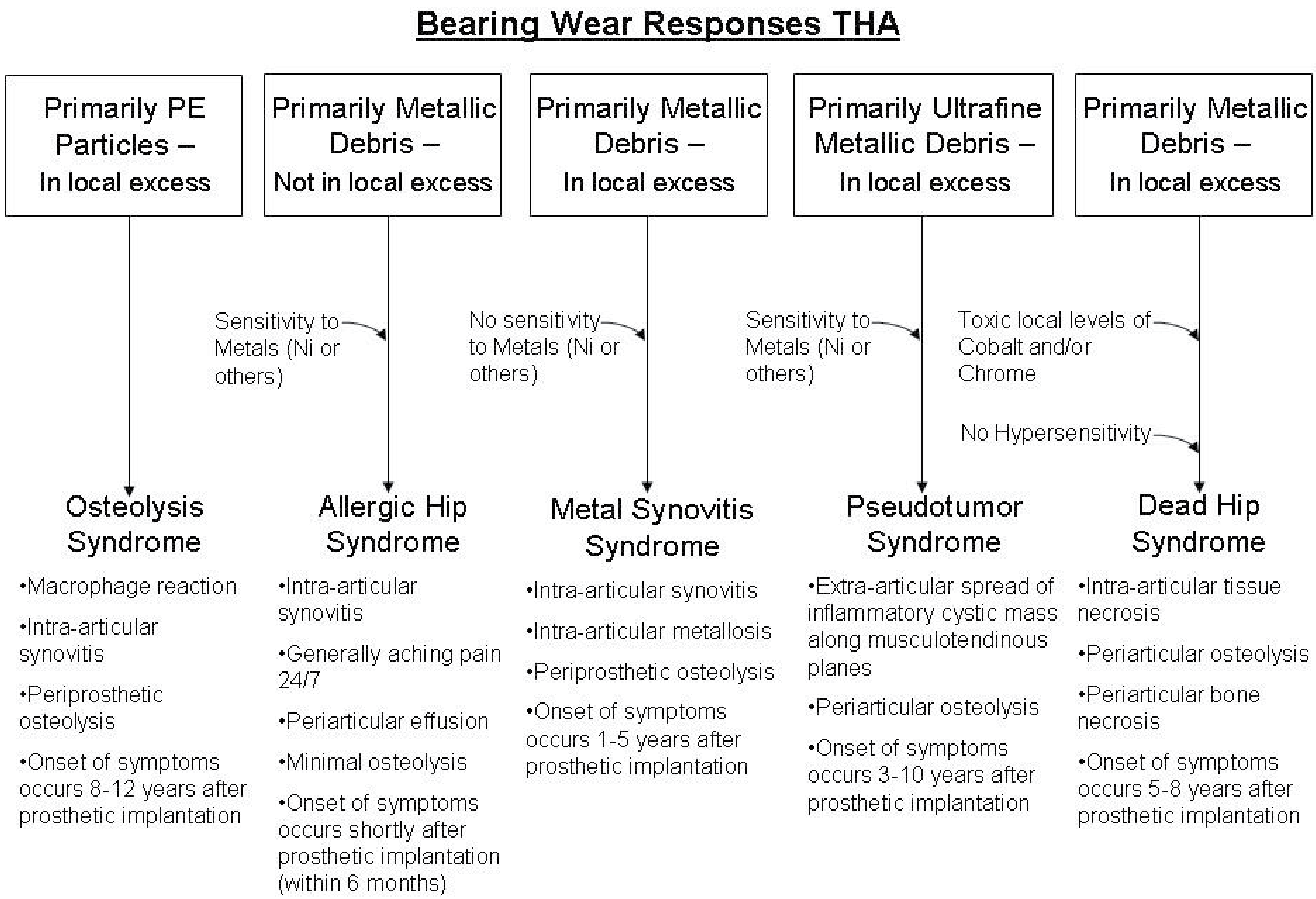


***Figure 10**. Diagram describing the proposed five major biologic wear responses in THA.*

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