



# Intrapelvic Pseudotumor Causing Neuropathy and Vascular Obstruction After Revision Total Hip Arthroplasty: A Case Report

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## Abstract

**Background:** There is a growing body of recent literature regarding the occurrence of pseudotumors associated with modular junctions and various bearing surfaces after total hip arthroplasty (THA). Revision surgery is often technically challenging and high complication rates have been reported. The optimal management of these patients and outcomes after operative treatment remain poorly understood.

**Methods:** We report the case of a 77-year-old male with progressive unilateral lower extremity swelling, pain, and neuropathy 9 years after revision THA for polyethylene liner wear. Imaging and biopsy confirmed a massive intrapelvic pseudotumor exerting compressive effects. Radiographs demonstrated extensive femoral and pelvic osteolysis without evidence of component loosening. Debulking of the intrapelvic portion of the pseudotumor was performed via the lateral window of the ilioinguinal approach with component retention.

**Results:** Debulking of the intrapelvic mass resulted in resolution of symptoms. One year postoperatively the patient reported pain free ambulation using a walker and no recurrence of symptoms. Radiographs demonstrated stable THA components in comparison with preoperative films.

**Discussion and Conclusion:** This case demonstrates a rare finding of intrapelvic pseudotumor causing neurovascular compression after revision THA. Clinicians should

be aware of intrapelvic pseudotumor as a possible cause of limb swelling and neuropathy, and that debulking of the mass is a potential treatment option in the setting of well-fixed implants.

## Background

Adverse local tissue reaction (ALTR) is being increasingly encountered in the setting of failed total hip arthroplasty (THA). Recent literature has focused on increasing our understanding of the biologic mechanisms that induce ALTR, as well as summarizing available evidence regarding the diagnosis and management of patients with this problem, which is recognized to lead to poorer outcomes [1-4]. Briefly, ALTR is a pathological biological tissue response that occurs in association with a joint replacement resulting from chronic immune response to wear debris. ALTRs include variable amounts of osteolysis, tissue necrosis, fluid collection, and soft tissue masses. 'Pseudotumors' are a form of ALTR, referring to benign, aseptic soft tissue masses which develop in the vicinity of a THA, and may be cystic, solid, or both [4-7]. The term pseudotumor is commonly used in reference to metal-on-metal

**Keywords:** Pseudotumor; Total Hip Arthroplasty; THA; Pelvic Mass; Adverse Local Tissue Reaction; Femoral Nerve Neuropathy  
**Level of Evidence:** IV

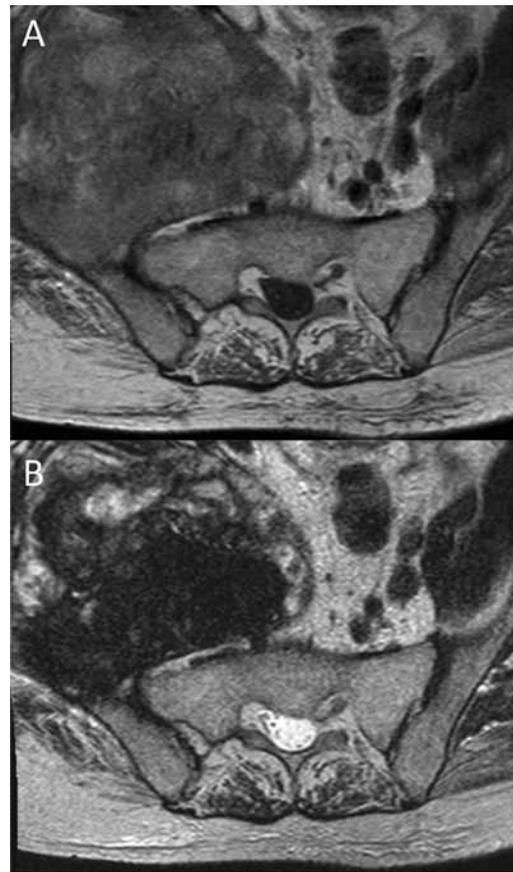
(MoM) THA, however, these masses have been associated with various types of THA bearing surfaces [1,4,5,8–11]. Pseudotumors most commonly present in the periarticular tissues and can be asymptomatic or may be a source of chronic pain due to soft tissue destruction or compressive effects. Here, we report the case of a patient with a large symptomatic pseudotumor with both intrapelvic and extrapelvic components after revision THA. The intrapelvic component resulted in vascular obstruction and femoral neuropathy, which successfully resolved after debulking of the mass. The patient was informed that his case would be submitted for publication, and he provided consent.

## Case Presentation

A 77-year-old male was referred to our institution for evaluation of right lower extremity edema, neuropathy, and pain associated with a right pelvic mass. The patient had previously undergone a ceramic-on-polyethylene primary THA in 1992 with a DePuy AML cementless femoral stem, ceramic femoral head, and Duraloc acetabular cup with a Hylamer liner (DePuy Orthopaedics Inc, Warsaw, IN). This was subsequently revised in 2010 by head and liner exchange for polyethylene wear and osteolysis. The acetabular liner was replaced with a Duraloc Marathon polyethylene acetabular liner (10 degree, +4 mm offset, 36 mm inner diameter, 56/68 mm outer diameter) and the ceramic head was revised to an M-Spec metal femoral head (36 mm diameter, 14/16 taper, +0 offset; DePuy Orthopaedics Inc, Warsaw, IN). Both the index and revision THAs were performed at outside institutions. He had no history of prosthetic joint infection.

The patient had a medical history of well-controlled diabetes mellitus (HbA1c <6.0), peripheral vascular disease, and atrial fibrillation managed with Coumadin. His pertinent surgical history included a right femoral-peroneal in-situ bypass graft, right L3-4 laminectomy, both within three years of presentation, and bilateral THAs. His body mass index was 26.9 kg/m<sup>2</sup> and he ambulated functional distances using a walker.

In early 2019, the patient began experiencing worsening buttock and groin pain, anterior thigh paresthesias, and diffuse swelling in the right lower extremity. His initial workup for radiculopathy was performed by his neurosurgeon. An MRI of the lumbar spine obtained incidentally revealed a large right-sided pelvic mass (Fig. 1). A computed tomography (CT) scan demonstrated a heterogenous mass in the right hemipelvis which measured approximately 13 x 10 x 26 cm in dimension (Fig. 2). A CT-guided biopsy was performed and revealed fibrinous and necrotic tissue



*Figure 1: Axial T1-weighted (Fig. 1-A) and T2-weighted (Fig. 1-B) MRI images of the pelvis demonstrating a large heterogenous mass in the right iliac fossa with compression of the pelvic cavity.*

with extensive histiocytic infiltrates and few foreign body giant cells. No organisms were cultured, no malignant cells were identified, and no metallic debris was seen. The patient was then referred to a regional academic medical center for evaluation by an orthopaedic oncologist. His case was presented at their multidisciplinary tumor board, and it was felt the mass was due to particulate disease related to his THA. He was then referred to our institution for definitive treatment.

Radiographs obtained during our initial evaluation demonstrated severe osteolysis involving both the proximal femur and acetabulum, with complete destruction of the superior pubic ramus (Fig. 3). The cementless stem and acetabular cup appeared well-fixed to bone with an intact superior rim and no obvious superior or medial migration of the cup. On physical examination, he had diffuse pitting edema throughout the right lower extremity to the thigh, and a mass-like fullness in the right gluteal region. He stated it felt as if he was “sitting on a tennis ball in the right hip.” There was no palpable mass over the iliac crest.

The mass had become very debilitating to the patient, and his primary complaint was significant swelling in the right lower extremity. Preoperatively, multiple lengthy discussions were held with the patient and his family on separate occasions. His imaging was reviewed, and the risks, benefits, and alternatives to both operative and non-opera-

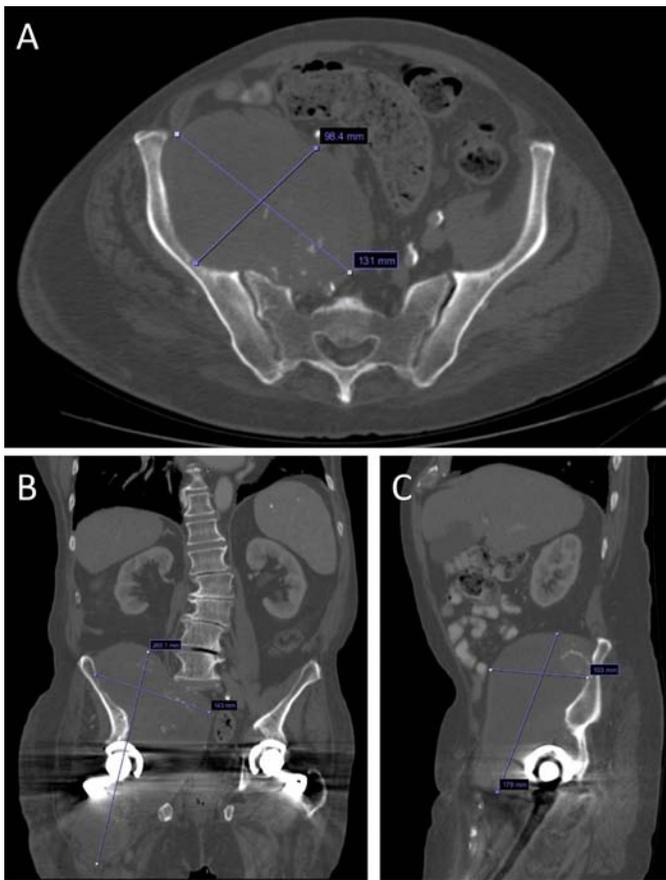
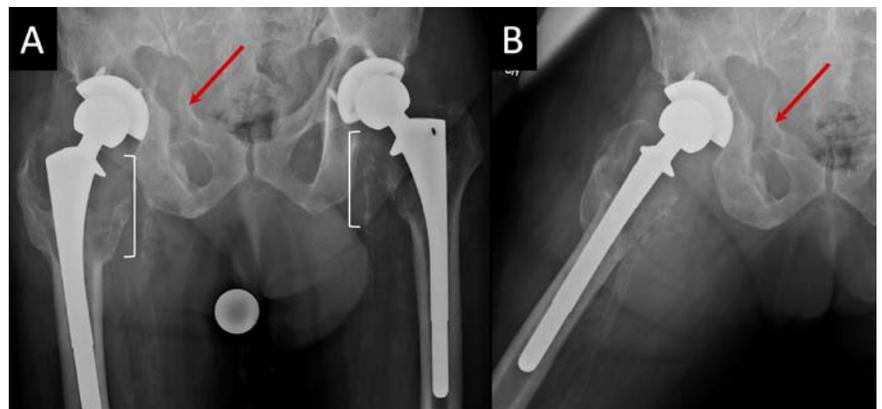


Figure 2: Preoperative axial (Fig. 2-A), coronal (Fig. 2-B), and sagittal (Fig. 2-C) CT images demonstrating a heterogeneous destructive mass with scattered peripheral and internal calcification measuring approximately 13 x 10 x 26 cm in dimension involving the right hemipelvis from the iliac fossa to midline. The lesion is causing mass effect on the intrapelvic structures and envelops the right acetabulum and hip prosthesis, extending into the posterior musculature of the right thigh. The mass communicates through a region of extensive osteolysis in the superior pubic ramus to the periprosthetic area.

tive treatment were thoroughly discussed. The complexity of his diagnosis and surgical options were explained, including the risks of progression of his osteolysis and further bone loss making acetabular and femoral reconstruction more challenging than his current situation. Furthermore,

Figure 3: Preoperative anteroposterior pelvis radiograph (Fig. 3-A) and frog-leg lateral radiograph (Fig. 3-B). There is extensive osteolysis involving both the proximal femur (demarcated in white brackets) and acetabulum on the right with less severe osteolysis on the left. There is complete destruction of the right superior pubic ramus (red arrows). There is an intact superior rim of the right acetabulum without obvious component loosening or migration.



his left THA, which also had significant radiographic osteolysis, was discussed. However, he was asymptomatic on the left side and deferred operative treatment. Ultimately, the patient clearly expressed his treatment goals, which entailed undergoing the least amount of surgery that could potentially alleviate some of his symptoms of swelling and paresthesia's. He had low functional demands and understood surgery would not be curative and there was a possibility of recurrence. After thorough discussion, he elected to proceed with debulking with the goal of relief of his compressive symptoms. Preoperatively, the decision was made to debulk only the intrapelvic portion of the mass, as it was felt to contribute to most of his symptoms. The case was reviewed with our vascular surgery colleagues who were available during the procedure.

In December 2019, the patient underwent debulking of the pseudotumor using the lateral window of the ilioinguinal approach. The anterior abdominal musculature was reflected medially off the iliac wing to expose an encapsulated mass adjacent to the iliacus muscle. This was incised, and more than 1L of thick, blood-tinged debris was evacuated (Fig. 4). The acetabular component was palpable



Figure 4: Intraoperative photograph taken through the lateral window demonstrating the porous coated shell visible through the inner table (white arrow) after debulking of the intrapelvic pseudotumor (Fig. 4-A). Clinical photograph of solid debris evacuated from within the fibrous wall of the mass (Fig. 4-B).

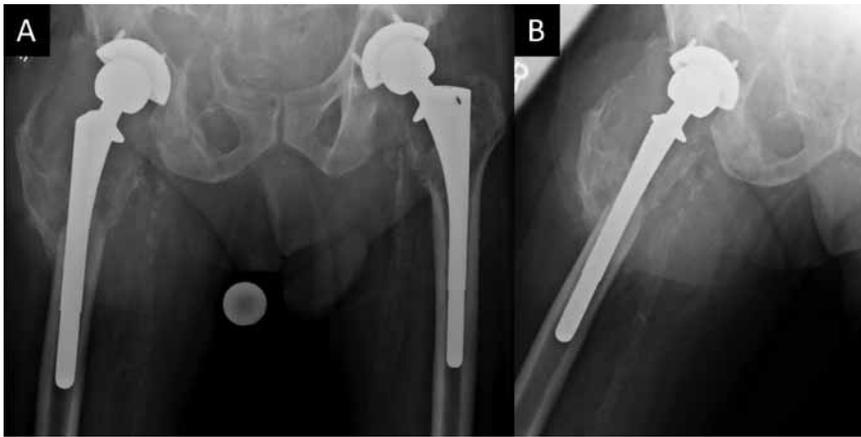


Figure 5: One-year postoperative anteroposterior pelvis radiograph (Fig. 5-A) and frog-leg lateral radiograph (Fig. 5-B).

through the cystic cavity and noted to be stable and well-fixed to bone. Due to the thickness of the debris, it was not possible to express further debris from within the leg. The patient had an uneventful recovery and was discharged to a rehabilitation facility.

One year postoperatively, the patient reported no pain or subjective limp, and was very pleased with the results of his surgery. His swelling had resolved, paresthesias were improved, and he was able to sit comfortably. He continued to use a walker for ambulation. Repeat radiographs demonstrated stable THA components in comparison with preoperative films (Fig. 5). He requested to defer any further operative treatment, unless his extrapelvic symptoms worsened.

## Discussion

Pseudotumor is an uncommon complication of THA, and the true prevalence is unknown [4,6]. In a meta-analysis, Wiley et al. reported a 0.6% estimated incidence of pseudotumor after MoM THA or resurfacing arthroplasty [12]. Higher prevalence has been reported in asymptomatic patients and after prolonged follow-up, suggesting growth of pseudotumors over time [13–16]. The rate of revision THA due to symptomatic pseudotumor has been reported to be 0.5% in non-MoM THAs and 2- to 3-fold higher in MoM THA [6,7,12,16,17]. Intrapelvic masses associated with a THA are rare, and have been reported in roughly 30 published cases [18–49]. Pseudotumors associated with THA may be asymptomatic, and identification may occur during routine investigations for another reason. While unexplained pain is typically the presenting symptom, unilateral limb swelling [25–27,29,45], sciatic nerve neuropathy [21], femoral nerve neuropathy [28,30,31], venous thrombosis [32–35], and ureteral obstruction [36,37] have been

reported in several case reports.

While the observed differences in biologic response to prosthetic debris vary between patients, particles of all types of metals, polyethylene, and ceramic debris have been shown to induce a biologic response and initiate osteolysis [50]. Our patient presented 27 years after his index THA and 9 years after his revision THA with extensive osteolysis and pseudotumor formation, which we suspect developed in response to wear debris related to his index THA liner. The accelerated wear, risk of osteolysis, and early failures of Hylamer liners, which were introduced in the early 1990s, are well-documented in the literature [51–54]. However, there is limited long-term follow-up information available on patients who have received this bearing surface. This data is important and an additional unique aspect of this patient's case worth highlighting.

The authors acknowledge several limitations associated with the perioperative workup for this case. The patient had undergone an extensive preoperative workup at several outside facilities prior to his evaluation in our office for definitive treatment, and thus a component of the decision making in this case relied on the quality of his medical records. Although the patient's clinical presentation, symptoms, outside laboratory studies, and previous biopsy results were not suggestive of a prosthetic joint infection, a full infectious workup would have ideally been performed at our institution. Additionally, while his pseudotumor and osteolysis were felt to be related to polyethylene wear, rather than metallosis, obtaining serum metal ion levels (cobalt and chromium) would have been prudent. Finally, unfortunately histological slides are discarded after a holding period at our institution and were not available to for image review of this case. Retaining a digital collection of intra-operative soft tissue specimens collected by institutions would be of value for patient care and retrospective case review.

Interestingly, our patient developed symptoms after previously undergoing a right sided lumbar spine decompression and right lower extremity vascular bypass. To our knowledge, this is the first case report to describe a pseudotumor in a patient with a history of operative spine and vascular surgery for symptoms in the ipsilateral extremity. The case presented also represents one of the largest pseudotumors documented in the literature.

The treatment decision for addressing symptomatic pseudotumors is challenging and options include aspiration, removal of the source of wear debris by component

revision or liner exchange, or resection with or without revision THA [3,4,26,39]. Bolognesi et al. provided a framework for the evaluation and treatment of patient with MoM THAs, and a recent consensus statement from the AAOS and AAHKS provides guidance for the evaluation and treatment of ALTR in metal-on-polyethylene THAs [2,7]. However, there is a paucity of data available to guide the evaluation and management of ALTR in patients who have previously been revised. We are aware of only three previous reports documenting enlargement or recurrence of a pseudotumor after revision THA and removal of the source of wear debris [31,55,56]. Revision THA for pseudotumor is recognized to have poor outcomes and high complication rates due to bone loss and periarticular soft tissue damage compromising stability [57–59].

To the best of our knowledge, there has been only one case reporting management of an intrapelvic pseudotumor with excision only [23]. However, in this case the patient refused a revision surgery and follow-up was not reported [23]. Most studies reporting intrapelvic pseudotumors document resection of the mass during revision THA, rather than for symptomatic treatment. In our case, debulking of the solid intrapelvic portion of the mass was performed to relieve compressive effects taking into careful consideration the patient's goals and functional demands. The decision to retain the acetabular cup was based on clinical and radiographic evaluation. The current case documents the potential for symptomatic relief after debulking of an intrapelvic pseudotumor.

In conclusion, clinicians need to be aware of pseudotumors as a differential diagnosis in patients with a history of THA who present with unilateral limb swelling, pain, or neuropathic symptoms. A multidisciplinary approach is advocated with close communication between musculoskeletal radiologists, musculoskeletal oncologists, pathologists, orthopaedic surgeons, and vascular surgeons. Debulking of the solid intrapelvic mass is a potential treatment option after revision THA in the setting of well-fixed components. Continued, long-term observation is warranted as re-accumulation of debris and recurrence of symptoms may occur.

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## SUBMISSION HISTORY

Submitted: June 28, 2021  
 Reviewed: September 10, 2021  
 Revised: September 15, 2021  
 Accepted: October 1, 2021  
 Published: October 12, 2021

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## AUTHOR DISCLOSURES

- The authors declare that there is no conflict of interest in connection with this submitted article.

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