

Giant cell tumor formation due to metallosis after open Latarjet and partial shoulder resurfacing

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Abstract

Shoulder metallosis with giant cell tumor formation is rarely seen in shoulder surgery. With an increase in shoulder arthroplasty and complex revision shoulder surgeries, clinicians should have an index of suspicion for possible metallosis in patients that presents with unexplained persistent pain with metal components on both the glenoid and humeral side. This case describes a 43-year-old female with a history of six prior shoulder surgeries who presented with shoulder metallosis and giant cell tumor formation after a screw from her open Latarjet procedure began rubbing against her Hemicap implant. She successfully underwent a revision total shoulder arthroplasty for post traumatic arthritis with pectoralis major transfer for her chronic subscapularis rupture and had complete symptom resolution.

Introduction

Metallosis is a known complication of metal-on-metal hip replacement that results from an increase in metal ions leading to a local inflammatory response. This reaction can cause aseptic loosening, granulation tissue formation, chronic pain, and joint failure.¹ Only a few case reports exist in the literature that describe metallosis in the shoulder, but there is no report on metallosis and giant cell tumor formation in the shoulder joint after multiple complex shoulder revision surgeries.²⁻⁵

We report a 43-year old female with a history of multiple revision surgeries done at outside hospital who presents to the

senior author (XL) with chronic right shoulder pain due to post traumatic arthritis and advanced metallosis with giant cell tumor formation of the soft tissue. We describe the patient's clinical presentation, management and a review of the literature about metallosis and giant cell tumor formation after shoulder surgery.

Case Report

A 43-year-old left-hand dominant female presented to our clinic with chronic right shoulder pain. She dislocated her right shoulder 12 years before and subsequently underwent 6 surgeries at an outside hospital, with the most recent being an open Latarjet procedure in 2011 and right hemicap resurfacing in 2013. She described pain with activity, rest, and at night. Her right shoulder subjective shoulder value (SSV) was 30% compared to her left which was 90%.

Neurovascular exam revealed 2+ radial and ulnar pulses with no sensory deficits. Right shoulder forward flexion was 0-100° actively and 0-170° passively. Abduction was 0-70° actively and 0-90° passively. External rotation was 0-100° passively on the right side, which was 40 degrees more than the contralateral normal side. Rotator cuff strength was 4/5 in forward flexion, 5/5 abduction, 5/5 external rotation, 4/5 belly press and 4/5 bear hug. There was no instability on apprehension and relocation test.

Plain right five-view (AP, Grashey, axillary, external rotation and scapular-Y) shoulder radiographs displayed two screws within the glenoid, humeral head resurfacing, glenohumeral osteophytes and osseous densities in the axillary recess (Figure 1A and 1B). MR arthrogram and CT showed a chronic full thickness subscapularis tendon tear, retraction to the level of the glenoid, grade IV Goutallier fatty infiltration, a large nonspecific joint effusion in the anterior inferior axillary recess and glenoid flattening with osteoarthritis. She had an ESR 13 mm/hr, CRP 5.4 mg/L and WBC of 10.9 K/uL. Ultrasound-guided aspiration was held for 3 weeks and was negative for any bacterial growth or P. Acnes.

The patient elected for a revision to total shoulder arthroplasty with pectoralis major transfer. The patient was positioned in the beach chair position under general anesthesia. Exam under anesthesia demonstrated forward flexion 0-170°, abduction 0-120° and external rotation 0-120° (60° greater than the contralateral

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side). Load and shift test were 2+ anterior, 1+ posterior and 1+ inferior. After sterile prepping and draping, a deltopectoral incision was made. Upon dissection, the subscapularis was absent and the capsule was thickened. The partial resurfacing was seen along with osteophytes on the humeral head (Figure 2A). A dark tissue was encountered within the glenohumeral joint (Figure 2B) and sent to pathology. The glenoid had exposed metal anchors which were subsequently removed. Additionally, there was humeral head medialization and medial wear on the glenoid exposing the shaft and one of the screws from the Latarjet procedure (Figure 2B). All hardware was removed and the patient was revised to a total shoulder arthroplasty (Figure 3A). The pectoralis major tendon was transferred just lateral to the lesser tuberosity (Figure 3B). Postoperatively, she was non-weight bearing in a sling with no external rotation past 20 degrees for 6 weeks. Standard postoperative physical therapy protocol was started. The two samples sent to surgical pathology were Specimen A (3.1x2.1x1.5cm tan-pink and tan-yellow nodule) which was a giant cell

tumor of the soft tissue and Specimen B (2.0x1.6x0.8cm tan-pink firm fragment) which was fibrous tissue and cartilage with degenerative changes. Histology H&E staining at 100x shows giant cell tumor formation (Figure 4) with the typical admixture of mononuclear cells, foamy macrophages (yellow star) and lymphocytes (orange circle) with scattered multinucleate giant cells (blue arrows).

At 1 year postoperatively, the patient reported significant functional improvement and had completed her formal course of physical therapy. Active forward flexion was 0-170°, active abduction was 0-90°, active external rotation was 0-60° with 5/5 strength in all planes of motion. She complained of mild apprehension although no instability was exhibited on load and shift test. Plain right five view shoulder radiographs were performed and reviewed consistent with appropriate postsurgical changes. One year postoperatively, her range of motion and strength exam from four months is unchanged. Her pain is much improved, and she is able to do her activities of daily living. Plain radiographs of the right shoulder were performed (Figure 5A and 5B). This patient was informed that her case would be submitted for publication as a case report and have provided her full consent.

Discussion

Metallosis is a well-known complication of metal-on-metal hip arthroplasty and is caused by elevated metal ion release from metal-on-metal contact which form immune protein complexes that can lead to pain, aseptic loosening, macroscopic necrosis, corrosive osteolysis, large sterile effusions and periprosthetic solid and cystic masses which fall under the spectrum of adverse reactions to metal debris (ARMD).⁶ Metallosis failure has been described in metal-on-metal hip arthroplasty, in addition to knee, elbow, and wrist literature. However, metallosis in the shoulder is rare (Supplementary Table S1). To our knowledge, this is the first case to describe metallosis leading to giant cell tumor formation of the soft tissue following open Latarjet and hemicap resurfacing due to friction between the two metal surfaces. As shoulder arthroplasty gains popularity, metallosis is a rare complication that should be considered in the differential diagnosis of patients presenting with unexplained pain in the setting of multiple revision complex surgeries and in patients with metal components on both the glenoid and

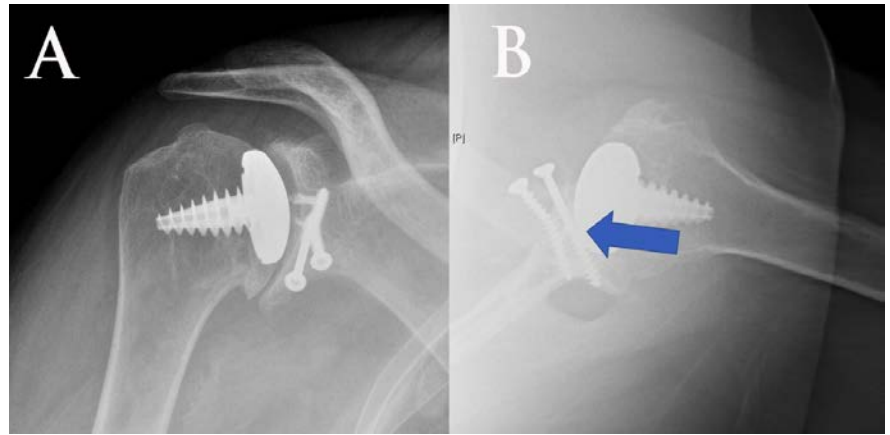


Figure 1. A. Right shoulder AP radiograph shows the partial resurfacing and two screws from the previous Latarjet surgery. There is also advanced stage glenohumeral arthritis. 1B. Axillary radiographic view of the right shoulder shows humeral head medialization and glenoid wear. The metal component of the partial shoulder replacement is in contact with the shaft of one of the screws from the Latarjet procedure (blue arrow).

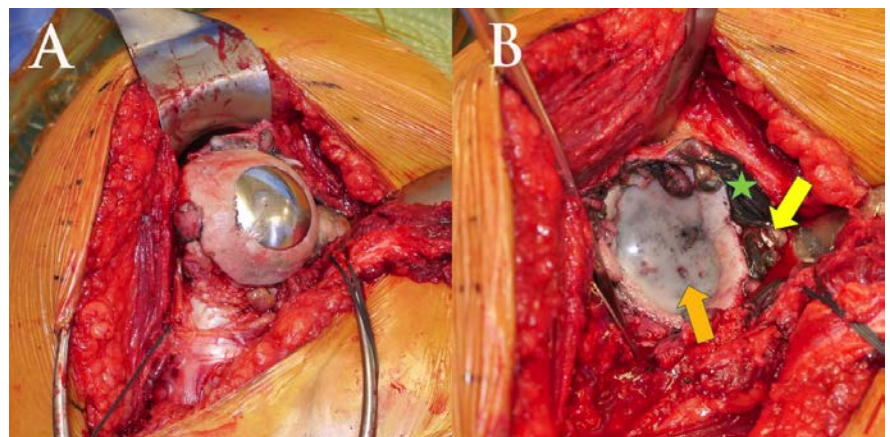


Figure 2. A. After exposure of the shoulder, partial shoulder resurfacing is seen with humeral head osteophytes. B. Dark tissue is above the anterior superior glenoid rim (green star) and the shaft of the screws is exposed (orange arrow) in the glenoid from the medial wear pattern. The yellow arrow shows the screw head.



Figure 3. A) Hardware removal and conversion to a total shoulder replacement. 3B. Full pectoralis major transfer is seen here with the blue arrow.

humeral side. The timeline for metallosis progression with giant cell tumor formation is variably reported. The earliest report of metallosis following total hip arthroplasty was 9 months, while in shoulder arthroplasty, metallosis has been reported as early as 28 months and as late as 8 years following primary surgery.^{5,7} Pseudotumor formation is a sequela of advanced stage metallosis and defined as a solid or cystic peri-prosthetic soft tissue mass with a diameter of >2cm that is not attributed to infection, malignancy, bursa or scar tissue and in the presence of giant cells on histology. Reports on symptomatic and asymptomatic pseudotumor prevalence in metal-on-metal hip arthroplasty has been reported between a range of 0.1-59%.⁸ Inflammatory pseudotumor etiology still remains poorly understood, but it is associated with a hypersensitivity reaction with development of periprosthetic cystic and solid or mixed masses.⁹ The timeline of pseudotumor formation in hip arthroplasty demonstrates an increase in incidence with prolonged follow up but is not clear about the amount of time needed for a pseudotumor to develop.¹⁰

A thorough history and physical examination are paramount to a structured workup for metallosis. Plain radiographs have demonstrated poor sensitivity for identifying metallosis and radiographic signs are absent in over half of the cases.¹¹ Routine plain radiographs should be obtained to assess the glenohumeral joint space, implant positioning, and osteolysis suggestive of loosening and fracture. Characteristic CT findings of metallosis are high-density enhancement outlining the joint capsule or bursa. MRI is the imaging modality of choice and findings are consistent with thin peripheral or septal enhancement, extension of the mass to the surface of the involved bone and mixed hyperintense and hypointense T2 signal.¹²

Differential diagnoses include infection and neoplasm. C-reactive protein (CRP) and erythrocyte sedimentation rate (ESR) tests, although nonspecific, are a useful and cost-effective method to rule out infection.¹³ In metallosis, serum markers of inflammation such as ESR and CRP are usually not elevated. Joint aspiration and cultures may assist in ruling out other etiologies including infection. In the case of pseudotumor, the aspirated fluid is typically black and this information may assist in making the diagnosis. Aspiration cultures will almost always be negative in cases of metallosis and giant cell pseudotumor.⁷ Serum metal ions of chromium and cobalt can be elevated with a threshold of 7ppb (particles per billion) and urine analysis

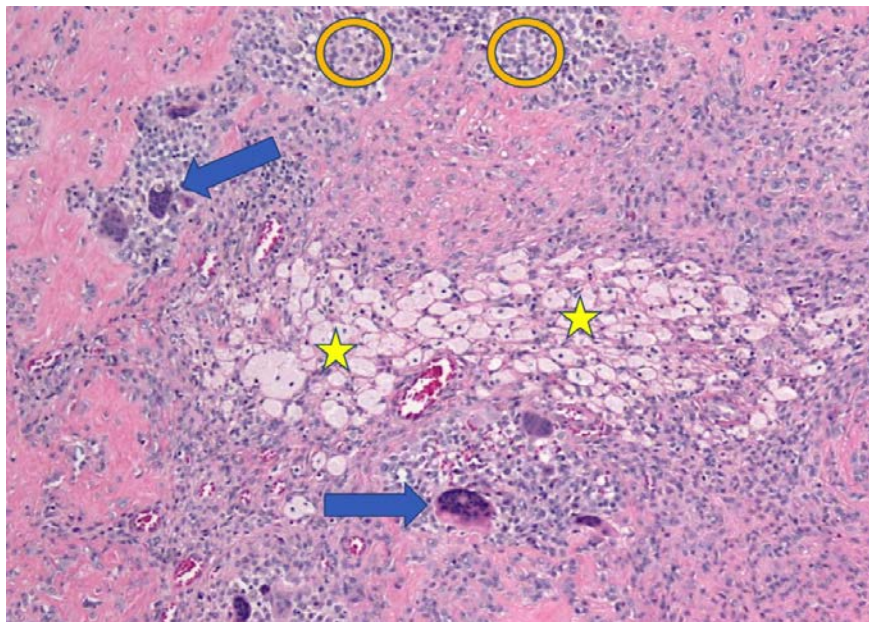


Figure 4. Histology H&E staining at 100x shows giant cell tumor formation in the soft tissue with the typical admixture of mononuclear cells, foamy macrophages (yellow star) and lymphocytes (orange circle) with scattered multinucleate giant cells (blue arrows).

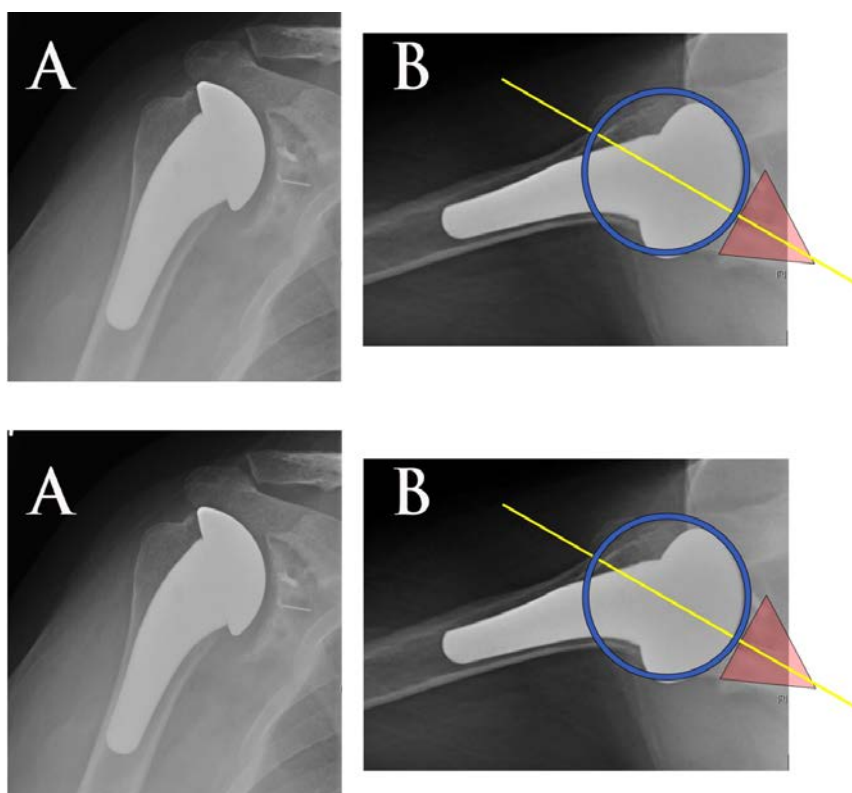


Figure 5. A. Post-operative AP radiograph at one year post surgery shows the total shoulder replacement. B. Axillary lateral radiograph shows the humeral head is well centered on the glenoid with no anterior subluxation detected.

may also reveal elevated metal ions.¹⁴ Metal sensitivity has been widely reported in the use of orthopaedic implants. Metallosis is considered to be a Type IV hypersensitivity reaction and skin testing may be considered during preoperative evaluation.¹⁵ Histologic findings have been reported from a macrophage response, metallic debris, foreign-body giant cells with black intracellular particles and necrotic fibrinous material with pigmented histiocytes and metallic debris.⁸ Intraoperative pathology is needed to confirm the diagnosis and rule out neoplasm. In our patient, the histology report documented the presence of typical admixture of mononuclear cells, foamy macrophages and lymphocytes with scattered multinucleate giant cells which is consistent with a pseudotumor reaction seen in the hip and knee literature in the setting of metallosis.

Conclusions

To our knowledge, this is the first reported case of advanced metallosis with giant cell tumor formation of the soft tissue in the shoulder after multiple complex revision surgeries. This complication was likely the result of the exposed metal shaft of her Latarjet screws and/or metal anchors from her prior surgery coming into contact with her metal hemiacap replacement leading to metal debris and chronic inflammatory giant cell reaction. As the prevalence of shoulder arthroplasty and revision surgery increases, metallosis may become a more common phenomenon. It is important to understand and recognize the

etiology of metallosis and giant cell tumor formation in the setting of shoulder surgery.

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