



## Case report

## Intramuscular hemangioma after total hip arthroplasty: an iatrogenic etiology

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## ABSTRACT

Soft-tissue hemangioma is a common benign tumor that can develop cutaneously, subcutaneously, or intramuscularly. Hemangioma formation within a muscular compartment is most often developmental in etiology; however, some cases are known to occur after blunt trauma to the soft tissues. To our knowledge, no cases of hemangioma formation after joint arthroplasty have been reported. We present a case of intramuscular hemangioma development within the hip abductor musculature after total hip arthroplasty via an anterolateral approach. Aside from developing congenitally or posttraumatically, hemangiomas may form after surgical dissection and must be considered as a source of anomalous swelling after surgery.

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## Introduction

Hemangiomas are common, benign proliferations of vascular-tissue origin. These tumors have a venous predilection and may occur within osseous or soft-tissue structures [1]. Soft-tissue hemangiomas exist cutaneously, subcutaneously, or intramuscularly [2]. The vast majority of soft-tissue hemangiomas are cutaneous in location with intramuscular hemangiomas, which makes up less than 1% of all hemangiomas [2]. Intramuscular hemangiomas are predominantly congenital in etiology with approximately 20% of cases occurring after trauma [3]. Most cases are reported in the pediatric and young adult population with approximately 90% of cases presenting before the age of 30 [3]. To date, there have been numerous reports describing intramuscular hemangioma developing congenitally, idiopathically [4], and posttraumatically [5]; however, to our knowledge, no cases of soft-tissue hemangiomas occurring after joint arthroplasty have been reported.

We present a case of a patient who underwent total hip arthroplasty (THA) for treatment of end-stage osteoarthritis and

subsequently developed a palpable mass localized over the left gluteal region. He was diagnosed with biopsy-proven intramuscular hemangioma within the hip abductor muscles and has been managed conservatively with a good overall functional outcome. The patient granted informed consent before preparation of this report.

## Case history

A 74-year-old man presented to an outside surgeon in 2013 with a 4 year history of progressively debilitating pain and stiffness localized to the left hip. Initial radiographs revealed significant left hip superior and medial joint space narrowing, subchondral sclerosis and osteophytosis consistent with severe osteoarthritis of the joint (Fig. 1). He was treated conservatively initially with activity modification, nonsteroidal antiinflammatory medication and ultrasound-guided left hip joint triamcinolone injection with mild symptomatic relief. He underwent a THA using a standard anterolateral approach [6] with detachment of the anterior one-third of the gluteus medius and minimus insertions from the greater trochanter. Using a standard technique, a size 54 Trident (Stryker Orthopaedics, Mahwah, NJ) acetabular shell and size 5 femoral stem were press-fitted into the reamed acetabulum and broached femur, respectively, without complications. The gluteal sleeve was repaired with a nonabsorbable, braided suture followed by a standard, layered closure with absorbable suture material.

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**Figure 1.** Initial preoperative anteroposterior (AP) pelvis radiograph.

His postoperative course was uncomplicated initially; however, roughly 3 years after undergoing primary THA, he began noticing a gradually enlarging, nonpainful, palpable mass over the left anterior gluteal region that increased in size with activity. A 10 × 5 cm nontender, fluctuant mass was noted along the lateral aspect of the iliac crest upon physical examination. The prior hip surgical wound was well healed without obvious signs of infection. Neurovascular examination of the operative extremity was otherwise unremarkable. Repeat radiographs demonstrated cementless acetabular and femoral components in acceptable overall alignment and position in addition to small islands bone formation within the gluteal musculature consistent with mature heterotopic ossification (Fig. 2). Progressive surgical-site swelling noted by the original surgeon prompted an magnetic resonance imaging (MRI) evaluation without gadolinium, which demonstrated a 10 × 7 × 10 cm multilobulated mass within the gluteal musculature that tracked toward the hip joint and into the subcutaneous fat superficially (Fig. 3). The mass was homogenous and dark on T1 and bright on T2 sequences. Laboratory studies included normal serum inflammatory markers (erythrocyte sedimentation rate 1 mm/h, C-reactive protein <0.4 mg/dL) and serum cobalt and chromium levels within normal limits (Co and Cr <1 micrograms/L). Given the abnormal appearance and signal intensity of the mass, the decision was made to proceed with open biopsy of the lesion by the senior author, an orthopaedic oncologist. A longitudinal incision was made over the gluteal musculature followed by blunt dissection towards the mass. The gluteus medius and minimus muscles were noted to be dusky and tan-brown in appearance with diffuse hypervascularity and overt bleeding; however, no fluid collection, purulence, or obvious masses were appreciated. A biopsy of the friable muscles was collected and sent for biopsy and culture. Thrombin-soaked sponges and electrocautery were used to achieve hemostasis, and the wound was closed in a layered fashion. Biopsy with hematoxylin and eosin staining demonstrated numerous vascular dilatations infiltrating skeletal muscle fibers and filled with erythrocytes (Figs. 4a and b). Immunohistochemical analysis demonstrated positive antibodies for desmin, CD31, CD34, and avian v-ets erythroblastosis virus E26 oncogene homolog (ERG), highlighting vascular proliferation in the setting of skeletal muscle (Fig. 5). No atypical cells suspicious for malignancy or infection were noted. This yielded a final diagnosis of intramuscular hemangioma.

Postoperatively, he developed a 3 × 2 cm superficial hematoma involving the biopsy site, which was treated conservatively with activity restriction. One month after open biopsy, he returned to baseline activities with complete regression of

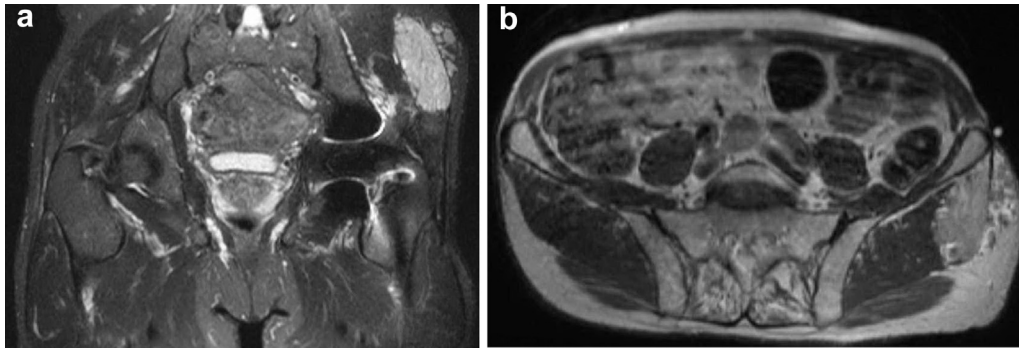


**Figure 2.** Postoperative AP hip radiographs.

the hematoma. The extensive involvement of the hemangioma in the gluteal musculature and lack of symptoms led us to pursue conservative management of the hemangioma with observation.

## Discussion

In our case, the occurrence of a mass after hip arthroplasty was confounding and expanded the differential diagnosis to include soft-tissue infection, adverse local tissue reactions (ALTRs), complex seroma or hematoma, synovial cyst, lymphatic or vascular malformation, and malignancy. The normal serum inflammatory markers and serum cobalt/chrome levels narrowed the differential and decreased the likelihood of acute infection and ALTR, respectively. Radiographs obtained after undergoing THA demonstrated calcified bone formation suspicious for heterotopic ossification. MRI showed a large mass with signal enhancement on T2 sequences consistent with fluid accumulation, benign neoplasm, or malignancy. The way in which the mass tracked between muscle groups and through fascia into the subcutaneous tissue was considered unusual for soft-tissue sarcoma. The MRI ordered before our evaluation did not include postgadolinium sequences making the diagnoses of hemangioma, arteriovenous malformation, and



**Figure 3.** Coronal short tau inversion recovery sequence (a). Axial T2 sequence (b).

malignancy such as angiosarcoma difficult to distinguish from hematoma, seroma, synovial cyst, or other fluid-containing lesions lacking blood flow. Although gadolinium would have better characterized the mass, the addition of contrast would have greatly enhanced the lesion's signal intensity, raising the suspicion of malignancy. Based on the appearance of the mass on MRI without contrast, the senior author, an orthopaedic oncologist, did not believe this would obviate the necessity of a formal biopsy. The remaining diagnoses, including malignancy and infection, were ruled out with open biopsy, immunohistochemical analysis, and cultures of the specimen.

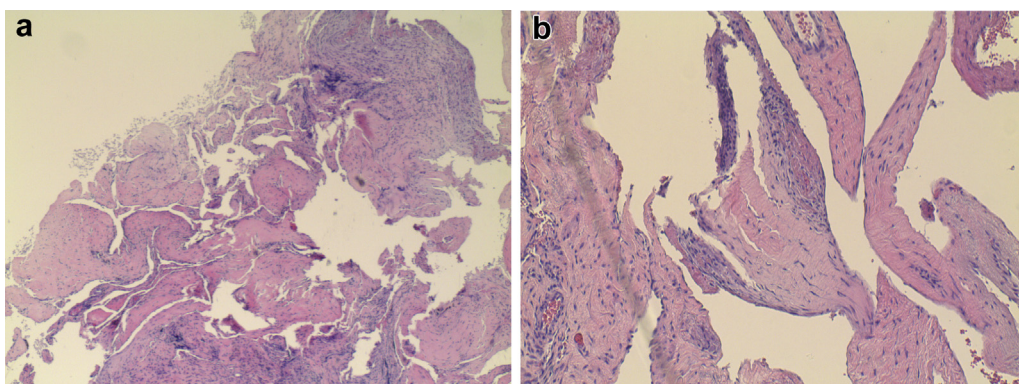
Given the tumor's pronounced size and intricate location within the gluteus medius and minimus muscles, surgical excision with marginal or wide resection would have led to extensive muscle debulking causing significant functional compromise. This, along with the patient's lack of significant symptoms and the high likelihood of recurrence, led to the decision to treat the lesion conservatively with observation.

Although the etiology of soft-tissue hemangiomas is primarily congenital or traumatic [4,5], this report brings to light the possibility that these tumors may arise after iatrogenic injury with surgical dissection. Patients with soft-tissue hemangiomas may present with symptoms of vascular engorgement including aching, pain, and swelling related to activity and lying in a dependent position [7]. Appropriate workup includes imaging modalities and laboratory analysis. Imaging studies for soft-tissue hemangioma evaluation may include plain radiography, ultrasonography, and MRI with the understanding that these neoplasms may resemble malignancy on cross-sectional imaging [8].

Although no specific laboratory studies exist in the diagnosis of hemangioma, the likelihood of acute infection and ALTR in the setting of THA secondary to corrosion at the metal head-neck junction may be lessened with normal serum white blood cell count, erythrocyte sedimentation rate, C-reactive protein, and serum cobalt and chrome levels [9].

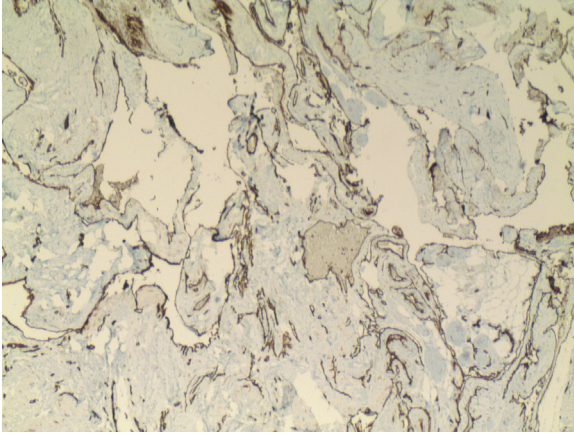
Treatment of soft-tissue hemangiomas has been a topic of controversy because surgical resection does not guarantee definitive resolution of the tumor and symptoms [10]. Bella et al [11] commented on recurrence rates in 110 patients who underwent surgical excision based on the degree of resection. Patients who underwent marginal or wide excision had a mere 7% recurrence rate, whereas patients undergoing intralesional excisions subsequently developed recurrence in 35%–67% of cases. Tang et al [10] reviewed 89 patients who underwent surgical excision with intralesional or marginal margins for soft-tissue hemangiomas. Authors concluded that surgical excision provided good pain relief and reliable outcomes. Boyd et al [12] commented on their outcomes after early surgical management of soft-tissue hemangiomas in 115 pediatric patients and concluded that excision, when done early, yields acceptable cosmetic and functional results.

In all, a hemangioma was revealed within the gluteal compartment 3 years after undergoing ipsilateral THA. We are unaware of prior reports in the literature of hemangioma development after undergoing surgery making this the first report of hemangioma arising from an iatrogenic cause. We believe that the trauma imposed on the periarticular vasculature at the time of surgical dissection may have led to cellular events, inducing neoplastic proliferation of injured endothelia.



**Figure 4.** Low-power micrograph of hematoxylin and eosin staining (a). High-power micrograph of hematoxylin and eosin staining (b).





**Figure 5.** Immunohistochemical analysis with anti-CD31 antibody, highlighting vascular proliferation.

### Summary

This case proposes an expansion in the etiology of hemangiomas highlighting the possibility that hemangiomas may occur after iatrogenic injury to the vasculature at the time of surgical dissection. Although a rare finding, we encourage iatrogenic hemangioma formation to be a part of the differential diagnosis in the patient

presenting with progressive, unusual postoperative swelling after undergoing surgery.

### References

- [1] Cohen AJ, Youkey JR, Clagett GP, Huggins M, Nadalo L, d'Avis JC. Intramuscular hemangioma. *JAMA* 1983;249(19):2680.
- [2] Mayerson JL, Scharschmidt TJ, Lewis VO, Morris CD. Diagnosis and management of soft-tissue masses. *J Am Acad Orthop Surg* 2014;22(11):742.
- [3] Watson WL, McCarthy WD. Blood and lymph vessel tumors. A report of 1056 cases. *Surg Gynecol Obstet* 1940;71:569.
- [4] Brown RA, Crichton K, Malouf GM. Intramuscular haemangioma of the thigh in a basketball player. *Br J Sports Med* 2004;38:346.
- [5] Patnaik S, Kumar P, Nayak B, Mohapatra N. Intramuscular arteriovenous hemangioma of thigh: a case report and review of literature. *J Orthop Case Rep* 2016;6(5):20.
- [6] Burwell HN, Scott D. A lateral intermuscular approach to the hip joint for replacement of the femoral head by a prosthesis. *J Bone Joint Surg Br* 1954;36-B(1):104.
- [7] Wild AT, Raab P, Krauspe R. Hemangioma of skeletal muscle. *Arch Orthop Trauma Surg* 2000;120(3-4):139.
- [8] Teo EL, Strouse PJ, Hernandez RJ. MR imaging differentiation of soft tissue hemangiomas from malignant soft-tissue masses. *AJR Am J Roentgenol* 2000;174:1623.
- [9] Fillingham YA, Della Valle CJ, Bohl DD, Kelly MP, Hall DJ, Pourzal R, Jacobs JJ. Serum metal levels for diagnosis of adverse local tissue reactions secondary to corrosion in metal-on-polyethylene total hip arthroplasty. *J Arthroplasty* 2017;32(9S):S272.
- [10] Tang P, Hornicek FJ, Gebhardt MC, Cates J, Mankin HJ. Surgical treatment of hemangiomas of soft tissue. *Clin Orthop Relat Res* 2002;399:205.
- [11] Bella G, Manivel J, Thompson R, Clohisy D, Cheng E. Intramuscular hemangioma: recurrence risk related to surgical margins. *Clin Orthop Relat Res* 2007;459:186.
- [12] Boyd VC, Bui D, Naik B, Levy ML, Hicks MJ, Hollier L. Surgery: the treatment of choice for hemangiomas. *Semin Plast Surg* 2006;20(3):163.