

Case Report

Spontaneous Rupture of an Inferior Gluteal Artery Pseudoaneurysm: A Case Report

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Gluteal artery pseudoaneurysms are rare, and most of the cases are due to blunt or penetrating trauma. Pseudoaneurysms most frequently involve the superior branch among all branches of the gluteal arteries, while involvement of the inferior gluteal artery is less commonly reported in the literature. Non-traumatic gluteal artery pseudoaneurysms caused by infection, inflammation, or other etiologies are uncommon clinical entities and may be easily misdiagnosed. Herein, we present a case of spontaneous rupture of an inferior gluteal artery pseudoaneurysm. Relevant literature on this topic was also reviewed.

Key words: pseudoaneurysm, inferior gluteal artery, embolization

Introduction

Pseudoaneurysm may be formed by damage to arterial wall through penetrating or blunt trauma, inflammation, infection, or iatrogenic causes. Gluteal artery pseudoaneurysms represent less than 1% of all aneurysms, and are mainly caused by blunt or penetrating injuries.¹ Other less common etiological mechanisms include atheroma, arterial dysplasia, polyarteritis nodosa, mycotic aneurysms, and aneurysmal evolution of a persistent sciatic artery.² Although prompt diagnosis can be made in patients with the “classic signs” of

posttraumatic mass with thrills, bruits, and pulsations, non-specific clinical manifestation and lack of obvious trauma history may easily lead to misdiagnosis and improper treatment. The incision and drainage of such a lesion can be disastrous if an erroneous diagnosis is made. We described a patient with non-traumatic gluteal artery pseudoaneurysm formation with spontaneous rupture that was treated successfully with N-butyl cyanoacrylate (NBCA) glue embolization.

Case Report

A 43-year-old man came to the emergency

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Received: June 25, 2012

Accepted: October 22, 2012

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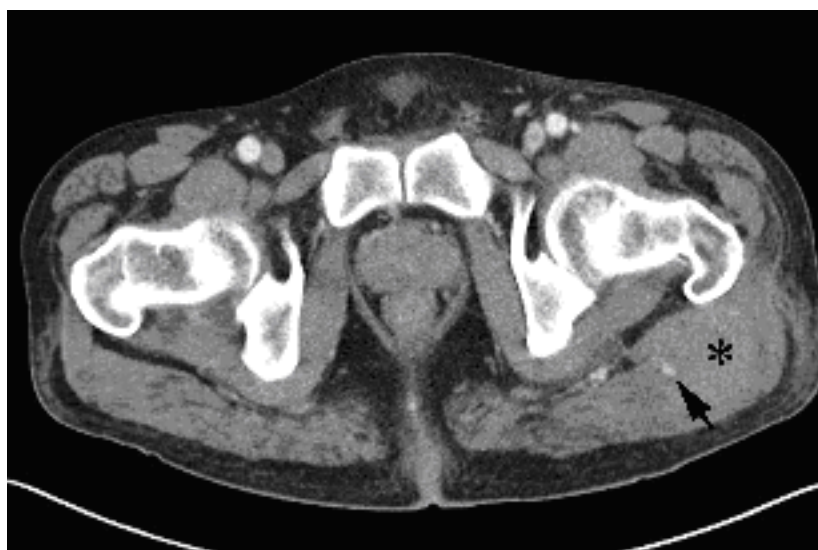


Fig. 1 Lower leg contrast enhanced-CT showing a contrast-enhanced focus (black arrow) with a hematoma in left gluteus muscle (asterisk).

department (ED) because of sudden onset of painful swelling and ecchymosis in his gluteal region and the posterior part of his left lower limb for three days. His past medical history included an appendectomy performed 30 years previously, gout, and daily alcohol use. He has no history of hypertension, diabetes mellitus, recent trauma or surgical procedures. The patient initially visited a local clinic for symptomatic relief of his left lower limb pain but without improvement. Due to progressive pain, ecchymosis, and swelling, he was then referred to our ED, a tertiary care center. On arrival, he was febrile with a body temperature of 38.2°C, a pulse rate of 80 beats per minute, a respiratory rate of 18 breaths per minute, blood pressure of 166/90 mmHg, oxygen saturation level of 99% at room air, and a Glasgow Coma Scale (GCS) of E₄V₅M₆. Initial physical examination showed a nonpulsatile swelling of left buttock and leg with tenderness and ecchymosis. Bilateral pedal pulses were present. Laboratory evaluation revealed an elevated white blood cell count of 13,970/ μ L with 76.0% of neutrophils, anemia with a hemoglobin value of 8.0 g/dL, a C-reactive protein concentration of 12 mg/L, electrolyte imbalance with a potassium level of 2.9 mEq/L, increased serum aspartate aminotransferase (AST) activity of

182 U/L, an elevated total bilirubin value of 2.91 mg/dL, a normal prothrombin time of 9.3 sec (INR: 0.90) and a platelet count of $302 \times 10^3/\mu$ L. Empirical antimicrobial therapy with intravenous ceftriaxone 2 g and clindamycin 600 mg were initiated under the tentative diagnosis of soft tissue infection. Computed tomography (CT) scan of the left lower limb demonstrated a contrast-enhanced focus with a hematoma in left gluteus muscle (Fig. 1) and another hematoma over left buttock, left semitendinosus and semimembranosus muscles. The imaging findings were considered suspicious for gluteal pseudoaneurysm. Two units of packed RBC were transfused and cardiovascular surgeon was consulted for left inferior gluteal artery pseudoaneurysm and an angiography was suggested. Through the right femoral artery, an angiography of the left internal iliac artery and its branches revealed a pseudoaneurysm at the branch of the left inferior gluteal artery (Fig. 2) for which transcatheter arterial embolization with NBCA glue was performed successfully.

The patient made a good recovery after embolization with substantial pain relief. Blood culture showed negative bacterial growth. He was discharged uneventfully 7 days after the procedure.

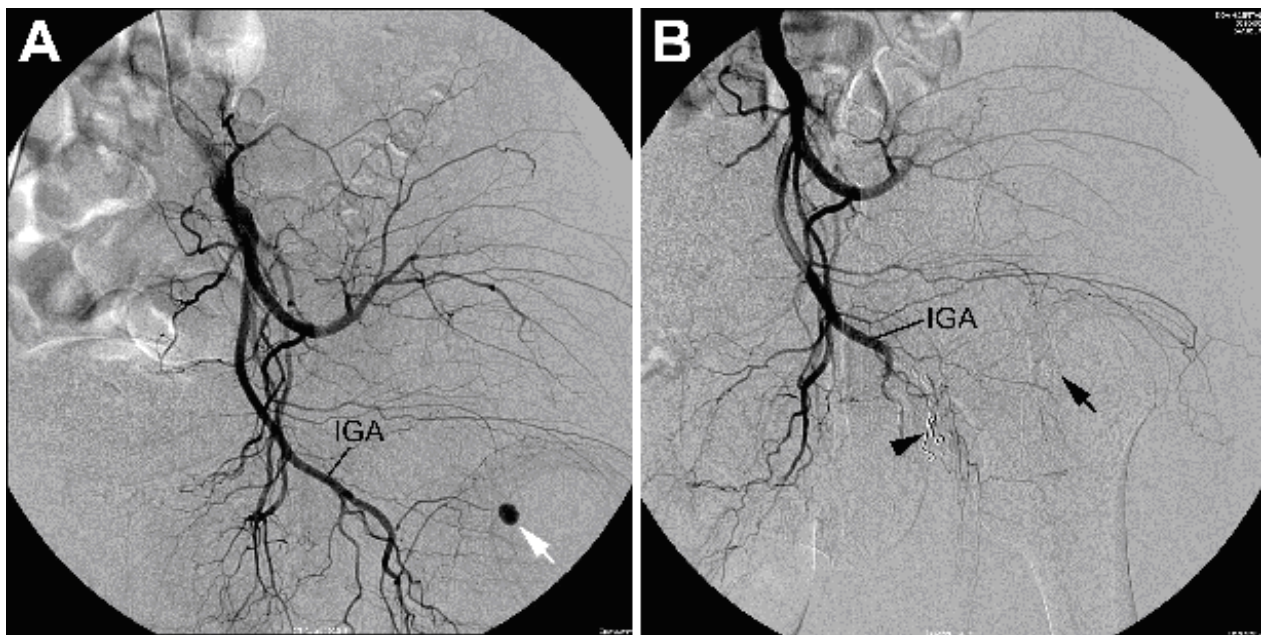


Fig. 2 Angiographic findings showing left inferior gluteal artery (IGA) pseudoaneurysm (A) before (white arrow) and (B) after (black arrow) NBCA embolization. The fibrocoils (black arrowhead) were deployed distally for protective purpose.

Discussion

A pseudoaneurysm is caused by disruption of an arterial wall, thus forming a sac that communicates with the artery. Gluteal artery aneurysms are rare, with the majority being secondary to blunt/penetrating trauma or iatrogenic causes. The superior gluteal artery is more commonly involved, while inferior gluteal artery pseudoaneurysms are less common with few cases reported in recent literature.³ The variable presentations, injury patterns, and symptoms may lead to misdiagnoses of these lesions. The aim of presenting this case is to highlight the importance of including pseudoaneurysm in the differential diagnosis when confronting a sudden-onset lower limb swelling in a non-traumatic patient, despite the absence of thrills, bruits, and pulsations.

A gluteal artery pseudoaneurysm can occur through several different mechanisms including penetrating or blunt trauma, intramuscular injections, muscle strain, inflammation or iatrogenic injury. Traumatic pseudoa-

neurysms often involve pelvic fractures and can present months or even years after the initial event.² Among patients with hereditary coagulopathy or those receiving anticoagulant therapy,⁴ trivial blunt trauma can lead to the formation of gluteal artery pseudoaneurysms. Infection of tuberculosis and methicillin-resistant *Staphylococcus aureus* (MRSA) may also cause gluteal artery pseudoaneurysms.⁵ A review of the literature demonstrates that the majority of patients with gluteal artery pseudoaneurysm are related to trauma, while sporadic cases have been described as complications of anticoagulant therapy.⁴ Since our patient presented with an abrupt onset of symptoms without trauma history, evidence of infection, and prior warning signs to indicate a pseudoaneurysm, making the diagnosis was even more challenging.

The clinical manifestations of a pseudoaneurysm vary widely. While it may be clinically silent, it may also present with local signs of pain and localized swelling due to mass effect or with systemic features including distal ischemia, embolization, or sepsis.⁶ The most fearful presentation is rupture, which

can lead to catastrophic consequences. Clinician should heighten his/her suspicion for a possible pseudoaneurysm when noting a firm mass after trauma. In addition to a pseudoaneurysm, differential diagnoses of a mass in the buttock area also include a pyogenic or tuberculous abscess, a soft tissue sarcoma, a lipoma, a hygroma, or an echinococcal cyst.⁷ Catastrophic consequences may result from attempts to perform drainage or biopsy of a soft tissue mass without careful evaluation. The presentations of a non-pulsatile swelling and ecchymosis of left buttock with extension to the posterior thigh and calf in our patient were different from the typical clinical manifestations described in the literature. Moreover, with such extensive involvement of the lower limb, it is important to include gluteal soft tissue infection with compartment syndrome and deep vein thrombosis in the list of differential diagnoses and to keep in mind that the compressive force can result in neurological impairment, muscle ischemia and necrosis.

The diagnostic imaging approach for pseudoaneurysms includes Doppler ultrasonography (US), contrast-enhanced CT, magnetic resonance imaging (MRI), and angiography. The first-line examinations include Doppler ultrasonography and contrast-enhanced CT, while angiography remains to be the gold standard for confirmation of pseudoaneurysm.⁸ Management options for peripheral artery pseudoaneurysm include surgery with arterial ligation, US-guided thrombin injection, endovascular or US-guided coil embolization, stent-graft placement, and endovascular glue embolization.⁹ The choice of treatment depends not only on the size, location, and neck width of the pseudoaneurysm, but also on the patient's clinical condition. Selective percutaneous embolization is now considered the mainstay of treatment in non-complicated gluteal artery aneurysms⁶ because of its lower risk of infection, the evasion of the retroperitoneal space, and the avoidance of the release of the tampon-

ade effect of the hematoma. If endovascular management fails or if the patient has signs of leg ischemia due to mass effect of the residual hematoma, persisting sciatic pain or sciatic nerve palsy, or has bladder or bowel dysfunction, open surgery should be considered to ligate the pseudoaneurysm and its parent artery, remove residual hematoma, or release compressed nerve tissue.¹⁰ Since our patient's general condition was relatively stable, he was treated successfully by NBCA glue embolization with excellent results.

Conclusions

While misdiagnoses of gluteal artery pseudoaneurysm may be attributed to their rare incidence and variable presentations, the diagnosis of spontaneous pseudoaneurysm rupture of unknown cause poses an even greater challenge to the emergency physician. Since the rupture of pseudoaneurysm or improper treatment may lead to devastating results, early and accurate diagnosis as well as proper management are crucial to improving patient outcomes. Emergency physicians should always include gluteal pseudoaneurysm in the list of differential diagnoses on encountering patients with trauma, risks of vascular injury, or coagulopathy presenting with swelling and ecchymosis of the lower extremity.

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