

A 69-Year-Old Man with a Painful Gluteal Mass due to a Superior Gluteal Artery Pseudoaneurysm Treated with Endovascular Repair

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ABSTRACT

Background: Arterial pseudoaneurysm, or false aneurysm, results from damage to the arterial wall, usually due to trauma or infection. Gluteal artery aneurysm is a rare condition with a frequency of less than 1% of all aneurysms. It occurs mainly as a complication of trauma. More seldom they are formed as a consequence of infections. The endovascular treatment is regarded as the gold standard. This report is of the case of a 69-year-old man with a painful gluteal mass due to a superior gluteal artery pseudoaneurysm treated with endovascular repair.

Case Report: A 69-year-old Caucasian male suffered from pain, feeling pressure and a pulsating mass in the left gluteal area for 6 months. There were no signs of infection in the laboratory tests. Magnetic resonance imaging (MRI) revealed a 35 mm contrast accumulating mass in the left superior gluteal artery (SGA). Scintigraphy with 99m-technetium-hexamethylpropylamineoxime (Tc-99m-HMPAO) labeled lymphocytes revealed the inflammatory character of the described abnormality. The pseudoaneurysm was secondary to endocarditis, which patients underwent one-year earlier. After cross-type catheterization of the right internal iliac artery digital subtraction angiography (DSA) revealed a 35mm pseudoaneurysm of the left superior gluteal artery. The mass was embolized distally and proximally. Control arteriography confirmed the effectiveness of the procedure. A control magnetic resonance angiography (angio-MR) study after three months proved complete pseudoaneurysm exclusion. Postoperative complications were not observed.

Conclusion: The endovascular treatment such as coil embolization of gluteal artery pseudoaneurysm is a safe and sufficient method, especially when performed on patients with many comorbidities e.g. as hypertension, heart insufficiency, and atrial fibrillation.

CASE REPORT

CASE REPORT

Arterial pseudoaneurysms are rare conditions, secondary to iatrogenic causes: complications of arterial access for endovascular procedures (incidence of <1%), anastomotic failure and non-iatrogenic causes: trauma, infection, pancreatitis with pseudocyst/pancreatic fistula. Treatment options of such pathologies include observation, ultrasound-guided compression, ultrasound-guided thrombin injection, and surgical repair: both open conventional and endovascular approach - treatment of choice [1].

Gluteal artery aneurysms occur with a frequency of less than 1% of all aneurysms [2-5]. There are only a few case reports where presented pathology was described.

It occurs mainly as complication of trauma e.g. traffic accidents [5-7]. Even more seldom they are formed as a consequence of polyarteritis nodosa, infections, dysplasia or persistent sciatic artery [8-11].

The process of diagnosis is often laborious as symptoms may occur long time after aneurysm occurs [3, 12].

In this study we report a case of successful diagnosis and treatment of mycotic pseudoaneurysm of superior gluteal artery, which was secondary to the endocarditis that patient underwent one year earlier, and successfully treated with endovascular approach.

CASE REPORT

A 69-year-old Caucasian male suffered from pain, feeling of pressure and pulsating mass in left gluteal area for 6 months before computed tomography (CT) (Figure 1). CT (December 2018) of the abdominopelvic cavity shows the pseudoaneurysm of the left SGA of 36x33mm. The patient suffered from many comorbidities as hypertension, heart insufficiency, atrial fibrillation treated with rivaroxaban. Moreover, he was treated for endocarditis caused by *Enterococcus faecalis* one-year previous to described procedure, he overwent biological aortic valve implantation, gastrointestinal stromal tumor resection, was treated for obstructive sleep apnea with continuous positive airway pressure, underwent hemithyroidectomy, had suspicion of rheumatoid arthritis and was planned for prostate cancer treatment. Additionally, he was overweight (BMI= 27,33 kg/m²), underwent appendectomy complicated by hernia, which was also treated surgically, suffered from multifocal demyelinating brain disorder of vascular origin, and essential tremor. After deep analysis the suspicion of the mycotic pseudoaneurysm in the left gluteal area was made.

He underwent CT in December 2018 which revealed 36mm contrast accumulating mass in the left superior gluteal artery and magnetic resonance imaging (MRI) in May 2019 confirmed each mass (Figure 1).

This finding was not present in previous scan in August 2018. Scintigraphy with Tc-99m-HMPAO labeled lymphocytes revealed inflammatory character of the described abnormality (Figure 2).

There were no laboratory signs of infection - C-reactive protein (CRP) and leukocytes levels were within norm. Due to his medical history suspicion of mycotic pseudoaneurysm was confirmed, because of previous endocarditis, clinical symptoms, and localization of the pseudoaneurysm. In view of recommendations endovascular procedure was performed.

After cross-type catheterization of the right internal iliac artery (which means that the puncture of the artery was on the left side of the body – into left femoral artery - and wire guide was introduced to the right common iliac artery through the bifurcation of the aorta, and next into the right internal iliac artery) digital subtraction angiography (DSA) revealed 35mm pseudoaneurysm of the left superior gluteal artery (SGA). The mass distally was embolised with two POD 15cm J-Soft Packing Coil, one 60 cm J-Soft Packing Coil (Penumbra Inc., Alameda, CA, USA), lipiodol, histoacryl and proximally by histoacryl and lipiodol. Control arteriography confirmed effectiveness of the procedure (Figure 3).

Puncture site has been secured by occlusive type system Angio-Seal 6F (Terumo, Somerset, NJ, USA). During the procedure 5000 units of the heparin were administered to protect patient against vascular complications. After the procedure the patient was stable, he was complaining about pain on the left lower limb. Analgesics were administered and gave patient relief of the pain. First and second day after procedure control morphology of the blood and CRP was in the range of norms. Antibiotics were not used in this case owing to any clinical symptoms (fever, erythema, oedema) and laboratory markers of inflammations. The patient was discharged 3 days after the undergone procedure.

Follow-up contains control angio-MRI study after 3 months, which proved complete pseudoaneurysm exclusion (Figure 4). Postoperative complications were not observed. Until now (42 months) after the procedure the patient remains under direct supervision of our department and has no signs of the excluded pseudoaneurysm.

DISCUSSION

Genesis or reasons of the disease

Gluteal artery aneurysms account for less than 1% of all aneurysms [2-5], with the superior gluteal artery affected more frequently than inferior gluteal artery [3, 13]. In comparison, abdominal aortic aneurysm has prevalence of 4 to 8 percent in screening studies [14-17].

There are only a few case reports where presented pathology was described. It occurs mainly as complication of trauma e.g. traffic accidents [5-7,15,16,29]. Gluteal artery aneurysms have incidentally been reported as a consequence of polyarteritis nodosa, mycotic infections, dysplasia or persistent sciatic artery [7-10].

The formation of pseudoaneurysms was reported following pelvic fractures (even years after initial injury) and after blunt injuries in patients with impaired haemostasis (anticoagulants, hereditary coagulopathies).

The complication after bone marrow biopsy is considered one of the most common causes of gluteal pseudoaneurysm. Other reasons include orthopedic surgery procedures (total hip arthroplasty, iliosacral screw fixation or iliac crest bone grafting), atherosclerosis, polyarteritis nodosa and intimomedial mucoid degeneration [18,24-28].

The diagnostic process is often laborious as symptoms may occur long time after aneurysm occurs [3,12].

The mycotic aneurysms are generally extremely rare pathologies, accounting for up to 3% of all aneurysms and diagnosed mainly in the aorta. They are observed in a significant percentage of patients who were diagnosed with endocarditis [13]. The mycotic aneurysm was first described by sir William

Osler in 1885 as a complication of the aforementioned endocarditis [29]. Up to this day only several cases of mycotic aneurysms in the gluteal arteries were described.

Fujimura et al. reports a case of the patient with mycotic aneurysm in superior mesenteric artery (SMA) and SGA secondary to *Staphylococcus epidermidis* endocarditis after mitral valve plasty. In their patients SMA aneurysm were treated conventionally while SGA aneurysm were embolized endovascularly [13].

Fiedler et al. presents a case of successful conventional treatment of a true non-typhi *Salmonella* mycotic aneurysm in a human immunodeficiency virus positive patient [30].

Akomea-Agyin et al. describes successful endovascular treatment of a superior gluteal artery pseudoaneurysm following aortic valve replacement because of *Streptococcus mitis* endocarditis [31].

Schindera et al. presents a results of a successful coil embolization of a methicillin-resistant *Staphylococcus aureus* (MRSA) SGA gluteal pseudoaneurysm in patient with a long history of drug abuse and perisacral abscesses due to chronic sacroilitis [32].

George N et al. presents a case of a successful endovascular embolization of a mycotic gluteal artery aneurysm secondary to *Streptococcus viridans* endocarditis [33].

Bouarhroum A et. al describes a case of 16-year-old boy with bilateral gluteal artery aneurysms associated with *Streptococcus mitis* endocarditis in whom left pseudoaneurysm was ruptured and treated using open surgery while contralateral one was successfully treated by elective embolization [34].

Bacterial endocarditis was also the reason of gluteal aneurysm (which was successfully embolized) in the patient of Grand et al. [10].

Our patient had the positive medical records in terms of risk factors of developing mycotic aneurysm. Having undergone mitral valve transplant and *Enterococcus faecalis* endocarditis, he was featured with high risk of developing mycotic aneurysm. It is worth mentioning that about 15% of patients undergoing bacterial endocarditis develop mycotic aneurysms, which in vast majority affect the aorta [31].

How to prepare accurate diagnosis?

The wide range of symptoms of the gluteal arteries aneurysms and a long time between the triggering disease and onset of the symptoms makes the diagnosis challenging and finding a connection between symptoms and previous medical history difficult. Gluteal artery aneurysms may present with localized pain, pressure, swelling and the sign of pulsating mass in the buttock area. The characteristic radicular pain, sometimes

with unilateral muscle weakness, mimicking the compression of spinal nerve roots on the level L4-L5 or L5-S1 were also described. Less specific symptoms as abdominal pain, nausea and vomiting, urinary or bowel dysfunction, and vascular insufficiency were also reported [7,22,29,33,35,37].

Our patient presented the most commonly described local symptoms: pain, pressure and pulsating mass in the gluteal area.

Doppler Ultrasound (DUS), Computed Tomography Angiography (CTA), MRI, as well as DSA can be used to diagnose gluteal artery aneurysm. Doppler Ultrasound can differentiate between false and true aneurysm, by visualizing characteristic turbulent flow and the connection between the neck of the aneurysm and the artery [1,7,35,36].

CTA and MRI may show the detailed anatomy and relations to the adjacent structures, while DSA may have both diagnostic and therapeutic character. Nuclear medicine methods (NM)– scintigraphy may, together with CTA and MRI, show inflammatory processes in the vicinity of the lesions indicating for mycotic character [18,26,34].

Our patient had the features of inflammatory lesions in CTA, which were confirmed by scintigraphy. The symptoms occurred more than year after endocarditis, therefore finding the connection between those two diseases was not obvious.

Proposal of treatment

The treatment options in case of pseudoaneurysms cover: open surgery, endovascular approach or hybrid procedures. If covered stent is not available or endovascular procedure cannot be performed it is possible to commence open surgery with using surgeon-reconstructed prosthetic graft and a bare metal stent [38].

Superior gluteal artery aneurysm can be treated endovascularly with using coil embolization [35].

Thrombin injection can be used alternatively to coil embolization; especially in acute superficial aneurysms with narrow necks [7]. It is potentially curative and decreases hemorrhagic complications if future surgical debulking is needed [36].

The endovascular treatment is regarded gold standard in management of gluteal arteries aneurysms with coil embolization considered the method of choice [24,33,34]. The first endovascular approach was performed by Herber et al. in 1988, who performed transcatheter coil embolization in order to decrease risk of bleeding during excising the aneurysm sac of the inferior gluteal artery [39].

In our patient the mycotic pseudoaneurysm was diagnosed. The treatment of the mycotic aneurysms includes antibiotic therapy accompanied by surgical tissue debridement and

vascular reconstruction. In such cases endovascular therapies are mainly reserved for patients with very high perioperative risk or ruptured aneurysms [40].

The first attempts of the treatment of mycotic aneurysms date back to 1924. Unfortunately, before the era of antibiotics vast majority were unsuccessful [41].

Contemporarily the endovascular treatment is regarded gold standard in management of gluteal arteries aneurysms with coil embolization considered the method of choice [27,42,43].

Before the era of endovascular methods, the open surgical repair was conducted. The aneurysms that were located intrapelvically, proximally to the sciatic foramen were operated with the difficult transperitoneal or retroperitoneal approaches, while posterior buttock approach was performed of extra pelvic lesions. Nowadays open surgical methods are introduced after endovascular attempt failure, or in case of complications which cannot be managed with minimally invasive techniques [7, 33, 34, 37]

Why should we choose minimally invasive techniques?

In this study the case of a 69-year-old man with a painful gluteal mass due to a superior gluteal artery mycotic pseudoaneurysm was reported. Because of the minimally invasive character of the endovascular treatment, as well as its suitability for high-risk patients with a large number of comorbidities, the endovascular method was chosen. Coil embolization was successfully performed with good short and long term results. This case shows that mycotic pseudoaneurysms may be difficult to be diagnosed and can be safely treated endovascularly with coil embolization.

CONCLUSIONS

Mycotic pseudoaneurysm of the gluteal artery is a rare condition that may appear in a long time after the occurrence of the predisposing factor in example – endocarditis. Nevertheless, key point of this case is to provided diagnostic properly: step-by-step and do not ignore any symptoms: such as pain, feeling of pressure in the gluteal area and correlate present symptoms with previous endocarditis (or other diseases which may cause pseudoaneurysms of gluteal arteries). Endovascular approach can be safe and effective method of treatment of this kind of pathology, featured with good long-term results.

DECLARATIONS

Ethical Approval and Consent to participate: Not applicable.

Consent for publication: Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Availability of supporting data: The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request. Figures' Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

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FIGURES

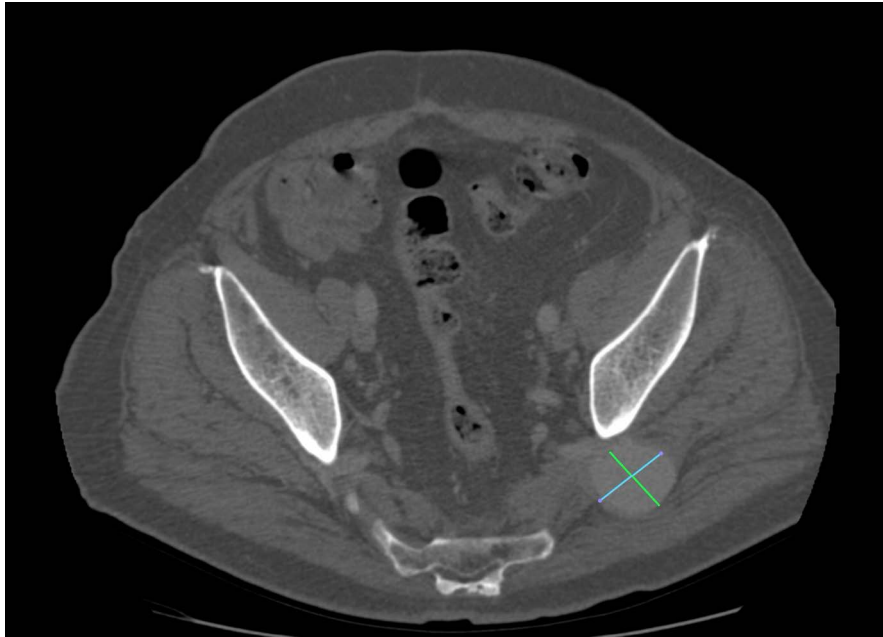


Figure 1: 69-year-old male with mycotic pseudoaneurysm of the left SGA. CT (December 2018) of the abdominopelvic cavity shows the pseudoaneurysm of the left SGA of 36x33mm.

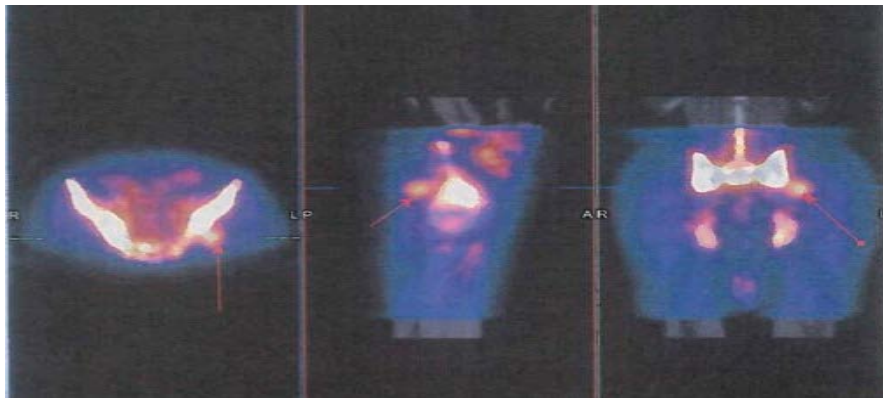


Figure 2: 69-year-old male with mycotic pseudoaneurysm of the left SGA. Scintigraphy with Tc-99m-HMPAO of pseudoaneurysm of the left SGA. Increased marked concentration – inflammatory character.

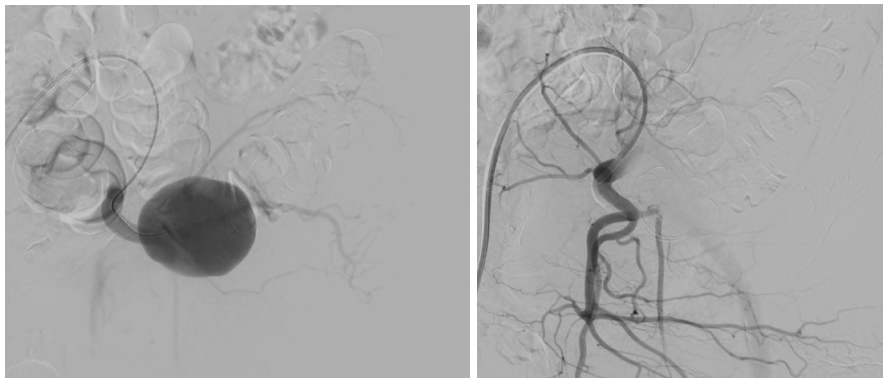


Figure 3: 69-year-old male with mycotic pseudoaneurysm of the left SGA. Arteriography (August 2019) before exclusion of the left SGA – A, and after exclusion – B.

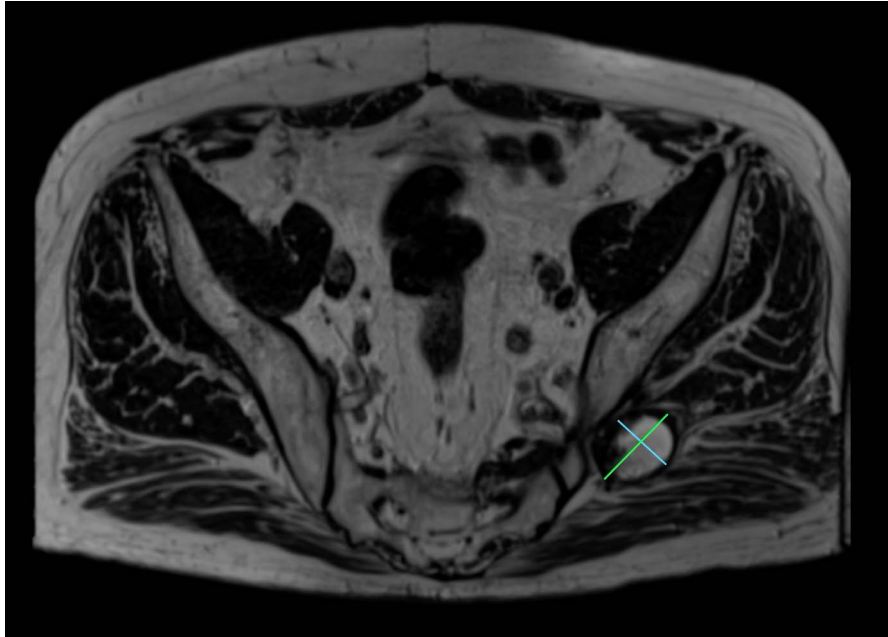


Figure 4: 69-year-old male with mycotic pseudoaneurysm of the left SGA. MRI (November 2019) after completed exclusion of pseudoaneurysm of the left superior gluteal artery 34 x 25mm.

KEYWORDS

Pseudoaneurysm; Gluteal Region; Endovascular Procedure

ABBREVIATIONS

MRI: MAGNETIC RESONANCE IMAGING
SGA = SUPERIOR GLUTEAL ARTERY
TC-99M-HMPAO = SCINTIGRAPHY
WITH 99M-TECHNETIUM-
HEXAMETHYLPROPYLAMINEOXIME
DSA = DIGITAL SUBTRACTION ANGIOGRAPHY
ANGIO-MR = MAGNETIC RESONANCE
ANGIOGRAPHY
CT = COMPUTED TOMOGRAPHY
CRP = C-REACTIVE PROTEIN
SMA = SUPERIOR MESENTERIC ARTERY
DUS = DOPPLER ULTRASOUND
CTA = COMPUTED TOMOGRAPHY ANGIOGRAPHY
NM = NUCLEAR MEDICINE METHODS
TEVAR = THORACIC ENDOVASCULAR AORTIC
REPAIR
EVAR = ENDOVASCULAR ANEURYSM REPAIR

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