

Large spontaneous median sacral artery pseudoaneurysm masquerading as hip pain

Joseph Faraj, MBChB, MS,^a Ali Daneshmand, MBChB, MS,^a and Bibombe Patrice Mwipatayi, MD, MMed, MCLinEd, FCS, FRACS,^{a,b,c,d} Perth, Western Australia, Australia

ABSTRACT

We report the case of an 81-year-old man who presented with severe right hip pain and reduced physical function secondary to a large spontaneous median sacral artery pseudoaneurysm measuring 83.5 × 55.4 mm. The patient had no history of recent trauma, infective or inflammatory disease, or any recent procedures. The patient was taking apixaban for atrial fibrillation. Percutaneous coil embolization was used to occlude the pseudoaneurysm. This case describes a novel disease, spontaneous median sacral artery pseudoaneurysm, presenting with hip pain. (*J Vasc Surg Cases and Innovative Techniques* 2020;6:224-7.)

Keywords: Pseudoaneurysm; Median sacral artery; Coils; Hip pain; Anticoagulation; Apixaban

Spontaneous pseudoaneurysm formation of the median sacral artery is an extremely rare vascular entity with catastrophic consequence if it is left untreated. Traditionally, arterial pseudoaneurysm formation is associated with recent trauma, infection, and inflammatory disease, or it can arise as a postoperative complication. Although there are a few reported cases of median sacral artery injury after a bone biopsy,^{1,2} formation of a large spontaneous pseudoaneurysm of the median sacral artery has yet to be described.

Both surgical and endovascular approaches can be employed in the treatment of a spontaneous pseudoaneurysm. Surgical exposure of the median sacral artery poses great technical challenges anatomically and is formidable to manage operatively. Transcatheter coil embolization is a well-established technique with demonstrated efficacy in treating similar pathologic processes, such as lumbar and visceral artery pseudoaneurysms, as described in the literature.³⁻⁵

This case describes the successful endovascular management of a novel disease and presentation—a spontaneous median sacral artery pseudoaneurysm that presented with the common complaint of hip pain. The patient consented to publication of this report.

CASE REPORT

An 81-year-old man with atrial fibrillation taking apixaban and with a medical history of chronic kidney disease (stage 3), hypertension, hypothyroidism, and obstructive sleep apnea presented to the emergency department with gradual-onset, severe right-sided hip pain when mobilizing during a period of 5 days. The patient reported no fevers or systemic symptoms. Furthermore, after further interrogation of the patient, he had a history of a fall that was not witnessed by any family member, but he did require help to stand up from the floor. He had no history of chronic back pain, previous bone biopsy, or back surgery. He was systemically well but reported having subjective fevers.

On examination, the patient's vital signs were within normal limits. His abdomen was soft and nontender in all quadrants as well as nontender over the bone landmarks of the anterior and posterior superior iliac spine. A targeted musculoskeletal examination revealed pain with right hip flexion and extension. There were no neurologic deficits. He had a routine peripheral vascular examination.

The patient underwent initial abdominal and pelvic radiography, which demonstrated no cause of his symptoms. However, computed tomography angiography of the aorta and iliac vessels revealed a large median sacral artery pseudoaneurysm embedded within the right psoas muscle measuring 83.5 × 55.4 mm (Fig 1). The full blood count was normal, with a white blood cell count of $6.5 \times 10^9/L$ and C-reactive protein level below 3.0 mg/L. All blood biochemical markers for an inflammatory or autoimmune process were negative. The hematology team was consulted for the reversal of apixaban in preparation for embolization of the pseudoaneurysm. His blood serum apixaban level was 312 ng/mL (reference range, 104.5-202.0 ng/mL), and he was subsequently administered 50 IU/kg of prothrombin complex (Prothrombinex; CSL Behring LLC, King of Prussia, Pa) for reversal.

The operation was performed under local anesthesia in a hybrid interventional operating room. A right femoral approach was chosen with a 5F access sheath. A pigtail flush catheter was introduced and placed in the aorta around the L3/L4 vertebra. The origin of the median sacral artery was not visualized on initial angiography. Rotational angiography was performed

From the Department of Vascular Surgery,^a and Medical Research Facility,^c Royal Perth Hospital; the School of Surgery, Faculty of Medicine, Dentistry and Health Sciences, University of Western Australia^b; and the Department of Vascular Surgery, Joondalup Health Campus.^d

Author conflict of interest: none.

Correspondence: Bibombe Patrice Mwipatayi, MD, MMed, MCLinEd, FCS, FRACS, Royal Perth Hospital, Department of Vascular Surgery, Level 2, MRF Bldg, 50 Murray St, Perth, Australia 6000 (e-mail: bibombe@iinet.net.au).

The editors and reviewers of this article have no relevant financial relationships to disclose per the Journal policy that requires reviewers to decline review of any manuscript for which they may have a conflict of interest.

2468-4287

© 2020 The Author(s). Published by Elsevier Inc. on behalf of Society for Vascular Surgery. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

<https://doi.org/10.1016/j.jvscit.2020.02.012>

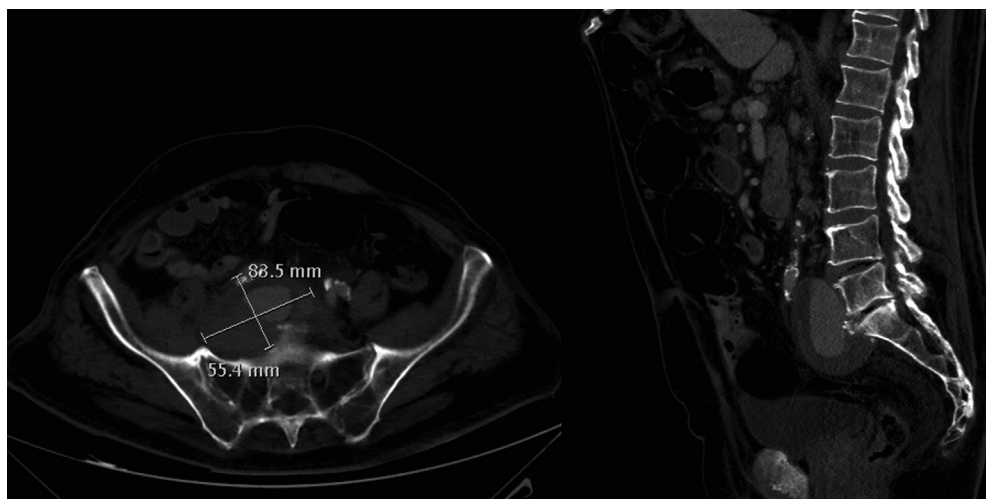


Fig 1. Computed tomography angiography image demonstrating the median sacral artery pseudoaneurysm in transverse and sagittal views with extension into the psoas muscle.



Fig 2. Computed tomography angiography and digital subtraction angiography images showing the median sacral artery (arrow) and partial filling of the pseudoaneurysm.

successfully and identified the median sacral artery with partial filling of the pseudoaneurysm (Fig 2). A 5 F MIK catheter was introduced over a guidewire and positioned at the ostium of the median sacral artery. The guidewire was exchanged for a 0.014-inch Asahi Chikai (Asahi Intecc Co Ltd, Aichi, Japan) neurovascular guidewire with a 2F Progreat microcatheter (Terumo Corp, Tokyo, Japan) introduced over the Asahi wire. The median sacral artery and the pseudoaneurysm were successfully cannulated. The Progreat microcatheter was then positioned into the pseudoaneurysm, which occluded inflow into the pseudoaneurysm. A decision was made to give 2 mL of thrombin (1000 units) before coil embolization in the event that embolization was not successful because of the instability of the working platform. Five 3- × 4-cm Concerto ev3 coils (Covidien, Irvine, Calif) were then deployed within the pseudoaneurysm, with the last coil deployed into the median sacral artery proximal to the pseudoaneurysm, occluding the inflow of the artery. Completion angiography demonstrated no flow through the median sacral artery or pseudoaneurysm. Closure of the right common

femoral artery access site was achieved with the FemoSeal vascular closure device (St. Jude Medical, St. Paul, Minn).

The patient was prescribed apixaban on day 2. There were no perioperative complications. He underwent a short period of rehabilitation and was discharged on day 15 from admission. Follow-up computed tomography angiography demonstrated successful occlusion of inflow to the median sacral artery pseudoaneurysm (Fig 3), and subsequently his right hip pain disappeared within 2 days.

DISCUSSION

Spontaneous median sacral artery pseudoaneurysms are extremely rare, such that we have been unable to identify a similar pathologic process in the literature. We are unaware of other cases of spontaneous median sacral artery pseudoaneurysms having presented with hip pain.

The median sacral artery is a small, single posterior branch that arises from the distal aorta before the aortic bifurcation. In some cases, it can arise from the fifth

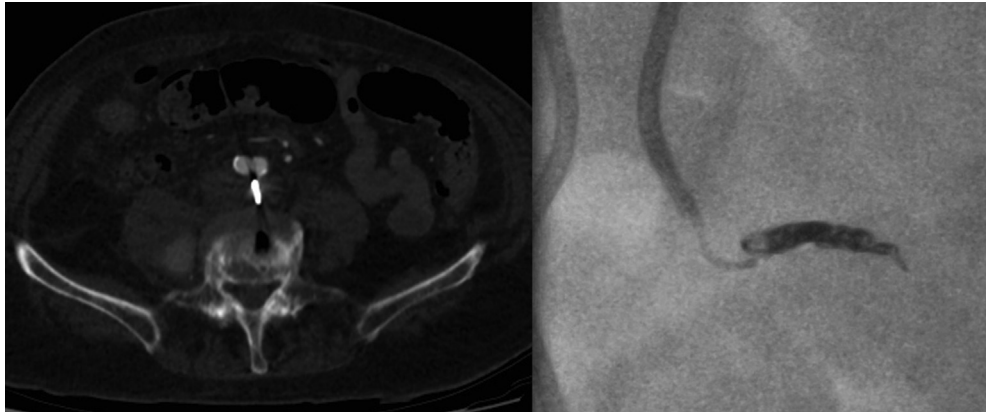


Fig 3. Postoperative computed tomography angiography and digital subtraction angiography images showing the coils in the sacral artery occluding inflow to the pseudoaneurysm.

Table. List of case reports on the formation of spontaneous pseudoaneurysms in the context of oral anticoagulation

Author	Year	Journal	Title	Anticoagulant
Guirgis et al	2017	European Journal of Vascular and Endovascular Surgery Short Reports	Spontaneous superior mesenteric artery branch pseudoaneurysm: a rare case report	Apixaban
Ikeda et al	2016	Journal of Medical Cases	Spontaneous rupture of a deep femoral pseudoaneurysm mimicking lymphedema after radical hysterectomy in a woman who was receiving warfarin	Warfarin
Kitamura and Mukohara	2014	Journal of Medical Case Reports	Spontaneous pseudoaneurysm of the hand	Apixaban

lumbar artery and can form a collateral circulation with branches of the internal iliac artery. It has anterior relations to the fourth and fifth lumbar vertebral bodies, posterior to the left common iliac vein, and is often accompanied by a paired vein.

Pseudoaneurysm formation in nearby vascular structures to the median sacral artery, such as the lumbar vessels, has been described in the literature mostly secondary to bone biopsy and vertebral body fracture. Pseudoaneurysms in this area may also arise spontaneously as a result of infective or inflammatory disease.⁵ However, our patient had no history of trauma, bone biopsy, or infective or inflammatory disease. The cause of the median sacral artery pseudoaneurysm was not determined; however, the patient's use of apixaban for atrial fibrillation was suspected to be a contributing factor.

It has been described in isolated case reports of a possible link between anticoagulation and the formation of spontaneous pseudoaneurysms⁶⁻⁸ (Table). It is postulated that individuals develop extravascular hematomas and recanalization of the hematoma, leading to formation of the pseudoaneurysm.⁷

Common presenting complaints for patients with pseudoaneurysms of nearby lumbosacral vessels are usually

back or flank pain. However, rupture of these pseudoaneurysms can occur, and patients may present in hemorrhagic shock requiring resuscitation and early operative management.⁹ An unusual presentation for pelvic pseudoaneurysms is hip pain and limited mobility mimicking a femur or pelvic fracture. Because of the age of the patient and the severity of the pain, initial thoughts were to exclude musculoskeletal disease; hence, workup for a femur neck fracture was performed.

CONCLUSIONS

Management can often be difficult and challenging. Surgery and endovascular methods are the two approaches that currently exist. Contrary to branched true aneurysms, pseudoaneurysms have a significantly higher rate of rupture (76.1% vs 3.1%) and must be treated immediately.^{9,10} The median sacral artery is in an anatomically difficult area and can be formidable to expose for the most skillful of surgeons. Surgical exposure can pose a serious risk to patients, specifically those who are fragile and have multiple comorbidities. It has been established that open repair of internal iliac artery aneurysms may result in serious adverse events, including buttock necrosis, iatrogenic venous bleeding,

and neurogenic complications.¹¹⁻¹³ Complications associated with pelvic aneurysms can be extrapolated to the open repair of median sacral artery aneurysms. Therefore, percutaneous endovascular coil embolization is an alternative treatment of choice with good outcomes and technical success for the treatment of pseudoaneurysm or hemorrhage of the lumbosacral vessels³⁻⁵ and management of median sacral artery pseudoaneurysm.

REFERENCES

1. Al Zahrani Y, Peck DJ. Median sacral artery injury following a bone marrow biopsy successfully treated with selective trans-arterial embolization: a case report. *J Med Case Rep* 2016;10:42.
2. Kim YU, Chae EY, Lee JH, Lee CH, Kim YK. Median sacral artery injury during percutaneous mechanical disc decompression using Dekompressor. *Korean J Anesthesiol* 2014;67(Suppl):S60-1.
3. Madhusudhan KS, Venkatesh HA, Gamanagatti S, Garg P, Srivastava DN. Interventional radiology in the management of visceral artery pseudoaneurysms: a review of techniques and embolic materials. *Korean J Radiol* 2014;17:351-63.
4. Belli AM, Markose G, Morgan R. The role of interventional radiology in the management of abdominal visceral artery aneurysms. *Cardiovasc Intervent Radiol* 2012;35:234-43.
5. Cordova AC, Sumpio BE. Visceral artery aneurysms and pseudoaneurysms—should they all be managed by endovascular techniques? *Ann Vasc Dis* 2013;6:687-93.
6. Guirgis M, Xu JH, Kaard A, Mwipatayi BP. Spontaneous superior mesenteric artery branch pseudoaneurysm: a rare case report. *EJVES Short Rep* 2017;37:1-4.
7. Kitamura A, Mukohara N. Spontaneous pseudoaneurysm of the hand. *Ann Vasc Surg* 2014;28:739.e1-3.
8. Ikeda S, Manabe T, Sugawara S, Sone M, Ishikawa M, Kato T. Spontaneous rupture of a deep femoral pseudoaneurysm mimicking lymphedema after radical hysterectomy in a woman who was receiving warfarin. *J Med Cases* 2016;7:299-302.
9. Abdelgabar A, d'Archambeau O, Maes J, Van den Brande F, Cools P, Rutsaert RR. Visceral artery pseudoaneurysms: two case reports and a review of the literature. *J Med Case Rep* 2017;11:126.
10. Hata S, Satoh R, Shin T, Mori K, Sumino Y, Satoh F, et al. Life-threatening rupture of an external iliac artery pseudoaneurysm caused by necrotizing fasciitis following laparoscopic radical cystectomy: a case report. *BMC Res Notes* 2014;7:290.
11. Soury P, Brisset D, Gigou F, Saliou C, Angel F, Laurian C. Aneurysms of the internal iliac artery: management strategy. *Ann Vasc Surg* 2001;15:321-5.
12. Rana MA, Kalra M, Oderich GS, de Grandis E, Gloviczki P, Duncan AA, et al. Outcomes of open and endovascular repair for ruptured and non-ruptured internal iliac artery aneurysms. *J Vasc Surg* 2014;59:634-44.
13. Stewart JR, Barth KH, Williams GM. Ruptured lumbar artery pseudoaneurysm: an unusual cause of retroperitoneal hemorrhage. *Surgery* 1983;93:592-4.

Submitted Oct 2, 2019; accepted Feb 23, 2020.