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CASE REPORT

# Spontaneous Superior Mesenteric Artery Branch Pseudoaneurysm: A Rare Case Report

**Mina Guirgis** [a], **Jema Hua Xu** [b], **Alar Kaard** [c], **Bibombe P. Mwipatayi** [d,e,*]

[a] Department of Vascular Surgery, Sir Charles Gairdner Hospital, Hospital Avenue, Nedlands, Perth, Australia
[b] Department of Vascular Surgery, Hollywood Private Hospital, Monash Ave, Nedlands, Perth, Australia
[c] Department of Radiology, Hollywood Private Hospital, Monash Ave, Nedlands, Perth, Australia
[d] Department of Vascular Surgery, Royal Perth Hospital, Wellington St, Perth, Australia
[e] School of Surgery, Faculty of Medicine, Dentistry and Health Sciences, Perth, Australia

**Background:** Visceral arterial pseudoaneurysms (VAPAs) are rare vascular entities with serious consequences. Traditionally, they are associated with trauma, infection, and inflammatory disease, or they can arise as a post-operative complication.

**Report:** An 87 year old man presented with abdominal pain and was found to have a spontaneous VAPA on a computed tomography angiogram. Serial imaging 4 months previously had demonstrated no aneurysm. Between scans, warfarin was changed to apixaban for aortic valve replacement, but he had no other changes to any other medications. He required urgent endovascular coiling of the pseudoaneurysm, with satisfactory recovery and outcome.

**Discussion:** VAPAs are extremely rare, with splenic artery VAPAs the most commonly reported. Regardless, fewer than 250 cases of splenic artery pseudoaneurysm have been reported. Superior mesenteric artery (SMA) pseudoaneurysms are the rarest type of VAPAs. Early identification and urgent treatment are warranted because of the associated high mortality risk, with a 50% risk of rupture in any given VAPA. Treatment options range from open operation to endoscopic and endovascular procedures. Apixaban has been proposed to contribute to pseudoaneurysm formation by slow and continuous bleeding that results in the formation of the pseudoaneurysm.

**Conclusions:** Spontaneous VAPAs are extremely rare and this is the first time a VAPA has been associated with the novel oral anticoagulant “apixaban”. Urgent management of any VAPAs is important because of the high risk of rupture and potential life threatening haemorrhage.



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## INTRODUCTION

Visceral arterial pseudoaneurysms (VAPAs) are rare vascular entities with serious consequences.[1] Traditionally, they are associated with trauma, infection, and inflammatory disease, or they arise as a post-operative complication.[2–4] Only one reported case of a spontaneous VAPA has been reported in the literature, involving the splenic artery.[5] Overall, there are fewer than a handful of reported cases of spontaneous pseudoaneurysms elsewhere in the body.

Although rare, VAPAs are of clinical significance because of their propensity to cause life threatening intra-abdominal or retroperitoneal haemorrhage.[5,6] Therefore, awareness of all aetiologies and prompt treatment are crucial for all VAPAs, whether they are symptomatic, haemorrhagic, or incidentally found.[5]

The first reported case of a spontaneous VAPA of the superior mesenteric artery (SMA) is described. Only one spontaneous VAPA has been reported previously in the literature, with the current case being the first to involve the SMA. Although rare, VAPAs are of clinical significance because of their propensity to cause life threatening haemorrhage. Therefore, awareness of their aetiology and prompt treatment are paramount.

## CASE REPORT

An 86 year old Caucasian man presented to the emergency department of the teaching hospital in April 2016 with exacerbation of intermittent abdominal pain and vomiting after a routine colonoscopy. His vital signs were within normal limits and he remained afebrile throughout. A

* Corresponding author. Department of Vascular Surgery, University of Western Australia, School of Surgery, Royal Perth Hospital, Level 2, MRF Building, Rear 50 Murray St, Perth 6000, Australia.
*E-mail address:* bibombe@iinet.net.au (Bibombe P. Mwipatayi).



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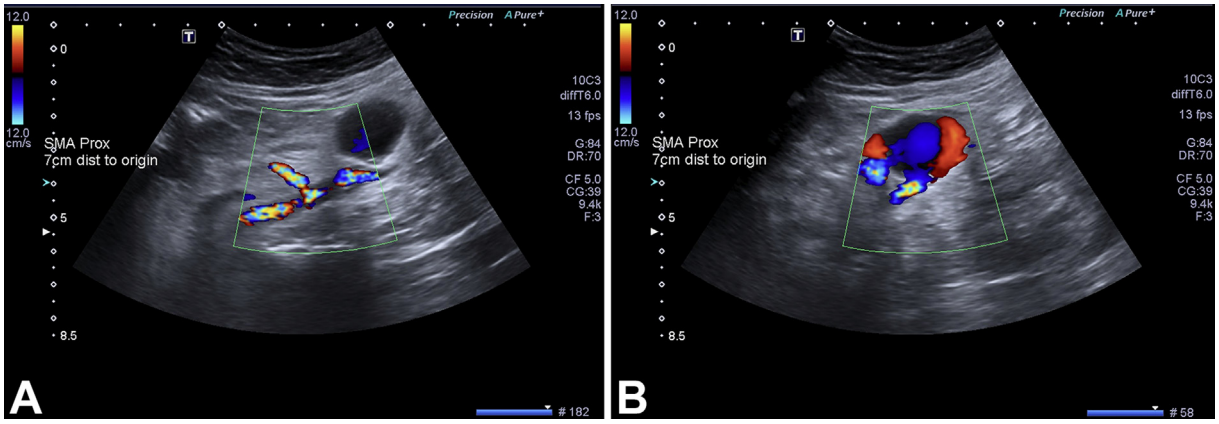


**Figure 1.** (A,B) Duplex scan demonstrate a high flow aneurysm within a proximal jejunal branch of the superior mesenteric artery. This measures 15 × 23 mm in diameter and contains only a small volume of mural thrombus. Prominent inflow and outflow vessels are identified.

mesenteric arterial Duplex scan demonstrated a high flow aneurysm within a proximal jejunal branch of the SMA, measuring 15 × 23 mm (antero-posterior [AP] × transverse [TV]) (Fig. 1A,B), and was followed by a computed tomography (CT) angiogram for work-up prior to any intervention. A CT scan performed 20 days previously because of abdominal discomfort demonstrated the same aneurysm measuring 19 × 15 mm (AP × TV), confirming a significant increase in aneurysmal size in the interim. His inflammatory markers, both white cell count and C-reactive protein, were not raised during his acute re-representation with abdominal pain, suggesting that the increased size of the VAPA was not associated with an inflammatory or infective process. Of note, just 4 months prior in December 2015, the patient presented with a right leg embolic event that was managed conservatively. The CT scan at that time did not demonstrate the presence of mesenteric vessel pseudoaneurysm but confirmed a high grade stenosis of the mid-femoral artery that was subsequently treated with a covered stent. The patient was discharged, and instructed to take apixaban 5 mg twice a day.

The patient had multiple comorbidities; in particular, a significant cardiac history, including congestive cardiac failure, a bioprosthetic aortic valve replacement, and recurrent pulmonary embolism, the last two warranting long-term anticoagulation therapy with apixaban. His non-cardiac medical history included type 2 diabetes mellitus, myasthenia gravis, and a left total hip replacement.

On discovery of this rapidly enlarging SMA pseudoaneurysm, semi-urgent endovascular repair was performed using a left brachial approach. A 90 cm, 6F Flexor Shuttle Guiding Sheath (Cook Medical, Bloomington, IN, USA) was introduced into the proximal portion of the SMA. A 100 cm Hinck catheter (Cook Medical) was inserted distally into the SMA. A 2.7 mm Progreat microcatheter (Terumo Medical Corp., Tokyo, Japan) was inserted into the Hinck catheter using a coaxial technique up to the jejunal aneurysmal sac, which was well visualised (Fig. 2A). The pseudoaneurysm and any branches feeding the aneurysm were coiled using an AZURCX and AZUR Framing Coil (Terumo Medical Corp.) (Fig. 2B).

The follow-up duplex scan of the mesenteric vessel at 14 weeks demonstrating normal flow in the SMA proximally

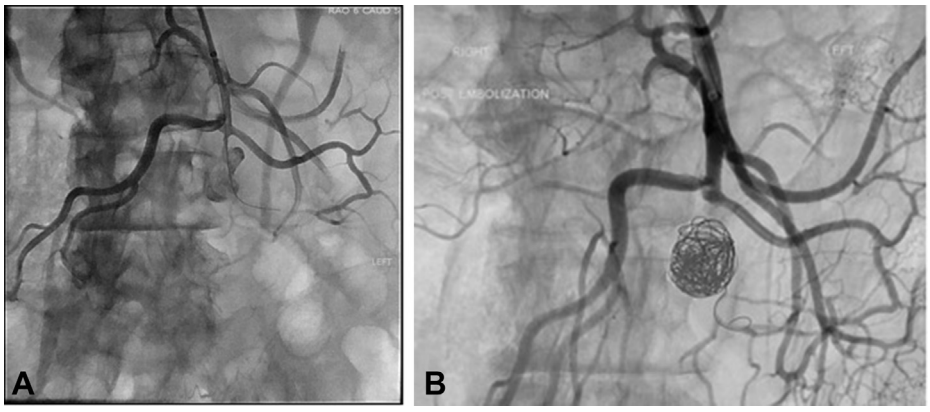


**Figure 2.** (A) Angiogram of the superior mesenteric vessel demonstrating the jejunal branch pseudoaneurysm identified by the contrast blush. The proximal superior mesenteric vessel; middle colic artery; ileocolic-right colic trunk; jejunal branch pseudoaneurysm; and jejunal arterial branch. (B) Angiogram post-pseudoaneurysm coiling showing no contrast blush.

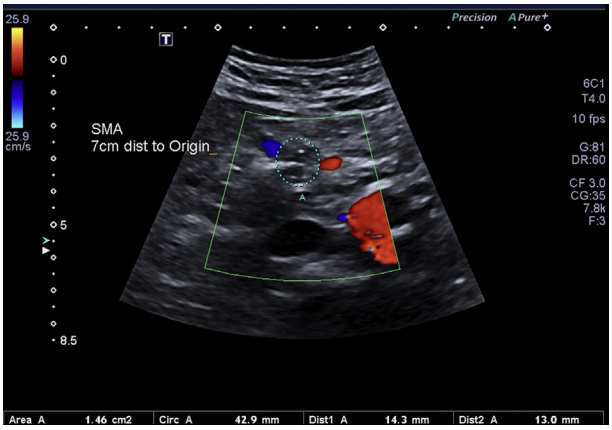


**Figure 3.** Duplex scan of the mesenteric vessel demonstrating a normal flow in superior mesenteric artery (SMA) proximally and distally. Embolisation material noted in SMA approximately 7 cm distal to the origin. There is no flow within this region to suggest recurrence of aneurysm.

and distally. Embolisation material was noted in SMA approximately 7 cm distal to the origin (Fig. 3).

There was evidence of satisfactory visceral perfusion with no evident ischaemic areas. The patient tolerated the procedure and recovered well, remaining pain free. Postoperatively, he was anticoagulated with therapeutic enoxaparin. Apixaban was ceased in the interim, as it was the only precipitating factor identified that potentially could have caused the spontaneous pseudoaneurysm. Ongoing surveillance for further aneurysmal events, with bi-yearly duplex scanning for the first 3—5 years, was scheduled on discharge. Written informed consent was obtained from the patient.

## DISCUSSION

Visceral pseudoaneurysms are extremely rare, with splenic artery VAPAs being those seen most commonly. Despite this, fewer than 250 reported cases of splenic artery pseudoaneurysms have been reported.[5] SMA and branches pseudoaneurysms are the rarest VAPAs, accounting for approximately 6—8% of all VAPAs, with an incidence as low as 0.01%.[1,7]

Despite being rare entities, early identification and urgent treatment are warranted because of the high mortality risk associated with them; there is a 50% risk of rupture in any given VAPA.[7] Treatment options range from open operation to endoscopic and endovascular procedures. Endovascular procedures include coil embolisation or covered stent placement.[8] Recently, there has been a shift towards endovascular coil embolisation, as it is associated with reduced morbidity and improved outcomes.[6]

The most commonly identified cause of SMA pseudoaneurysm is pancreatitis or trauma. The proposed mechanism of vascular injury in pancreatitis involves pancreatic autodigestion enzymes being released into the perivascular space, leading to enzymatic digestion of the arterial wall.[8] Rarer causes of SMA pseudoaneurysms include infective endocarditis or uncontrolled hypertension.[7,9] To date, there has been no published report of an idiopathic SMA pseudoaneurysm.

The only change in this patient's management was the recent commencement of apixaban for aortic valve replacement.

Apixaban, an oral anticoagulant, works by directly inhibiting both free and clot bound factor Xa, in turn preventing thrombin generation and further clotting.[10] In the setting of procedural complications, apixaban has been found to cause VAPA, but this is extremely rare.[11] Apixaban has not been found to cause a non-procedure related pseudoaneurysm in any major medical database. The commencement of apixaban in this patient is proposed to be related to the formation of his VAPA.

## CONCLUSION

Spontaneous visceral pseudoaneurysms are extremely rare; this case report seems to be the first account of a visceral pseudoaneurysm associated with a novel oral anticoagulant, which in this patient was apixaban. It is of utmost importance to urgently manage any VAPAs because of the high risk of rupture and potential life threatening haemorrhage. It is important to recognise that there is no obvious correlation between the pseudoaneurysm size and risk of rupture.[12] Among the options for treating VAPAs, endovascular coil embolisation is a safe and effective technique for definitive management, especially for poor surgical candidates.

## CONFLICT OF INTEREST

None.

## FUNDING

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