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Case Report

Spontaneous isolated superior mesenteric artery dissection: an investigative case report ^{☆,☆☆}

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ABSTRACT

Spontaneous isolated superior mesenteric artery dissection (SISMAD) is a rare cause of acute abdominal pain, but could potentially be fatal to patients, and should be recognized soon in the emergency department after excluding other common causes. Computed tomography (CT) is the modality of choice for initial diagnosis and follow-up. Currently there is no evidence-based guidelines for managing SISMAD. A 58-year-old man being suspected of a mesenteric artery dissection was referred to our emergency department. The patient was monitored, treated conservatively with anticoagulant and discharged after 3 days. Follow-up CT scans at 6 month, 1 year and 1 year and a half post discharge showed a partially occluded false lumen, the diameter of true lumen had increased in size and no signs of bowel ischemia. SISMAD should be considered as part of differential diagnoses when patients in their fifth to seventh decades of life present with acute abdominal pain. Treatment includes conservative management, percutaneous endovascular interventions, or surgery, but most patients can be managed conservatively.

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List of abbreviations: SISMAD, Spontaneous isolated superior mesenteric artery dissection; CT, Computed tomography; SMA, Superior mesenteric artery.

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Introduction

Spontaneous isolated superior mesenteric artery dissection (SISMAD) is a rare vascular disease first described in 1947 in an 87-year-old Caucasian woman who presented with intestinal obstruction [1]. Recently, the number of reports on SISMAD increased significantly, primarily due to the widespread availability of modern cross-sectional imaging modalities [2]. Nonetheless, the true incidence remains underestimated and underreported [2], with limited expertise, consensus, and evidence-based guidelines drawn for diagnosing and managing SISMAD.

The prevalence of SISMAD is difficult to estimate to its reporting as isolated case reports or case series, but multiple reports have shown a higher prevalence amongst males in their 50s-70s [3]. Additionally, there seems to be geographical predominance in Asian countries [3]. Herein, we describe the case of a 58-year-old man with SISMAD managed at our institution.

Case report

A 58-year-old man was suspected of mesenteric artery dissection based on abdominal CT scan at a low-resource setting hospital and was referred to our emergency department with only the CT scan report. He presented with chief complaint of severe acute abdominal pain that had lasted for a day, localized most notably to the periumbilical area and radiated toward his epigastric region. The pain had an abrupt onset while he was relaxing, and peaked almost instantly. On admission, the man was conscious with stable vital signs, seemed distressed and nauseous. He did not complain of diarrhea or bloody stools. There was no identifiable pain aggravating or alleviating factors. He denied any abdominal trauma.

The patient had well-controlled hypertension, but untreated dyslipidemia. He had a history of smoking 60 pack-year but stopped 10 years ago, and no history of peripheral artery disease, or diabetes.

There was tenderness over the periumbilical area on palpation. A digital rectal examination showed no blood. Otherwise, the examination was unremarkable. Neutrophil counts were at the upper normal level, and he had a C-reactive protein of 30.2 mg/dL (normal range, <5 mg/dL). The rest of the comprehensive metabolic and coagulation panel were usual. Contrast-enhanced thoracic-abdominal CT scan revealed an intimal flap in the superior mesenteric artery (SMA) and a partially occluded false lumen (Fig. 1). The true lumen was only 3 mm, but there were no signs of bowel ischemia.

He was treated conservatively with pain medication, fluid resuscitation, intravenous heparin, and close monitoring in the general surgery department with serial abdominal examinations, at least twice a day. The pain subsided, the man was discharged after 3 days. He continued warfarin at home with an INR goal of 2-3, and received follow-up appointments every month for the first 6 months after being discharged.

Six months postdischarge, a contrast-enhanced CT scan showed an occluded false lumen and there was less fat stranding around the SMA. Only at this point the entry point could be

located at 3.6 mm from the origin of SMA, its diameter was 2.4 mm (Fig. 2). There were still no signs of bowel ischemia (Fig. 3). From this point, he was followed-up every 3 months.

CT scans at 1 year and 1 and a half year postdischarge (Fig. 4) revealed that the thrombolysed false lumen decreased in size, and the diameter of SMA true lumen had increased to 5.2 mm and stayed relatively stable, with no signs of bowel ischemia.

Discussion

SISMAD remains the most common visceral artery dissection [3]. The risk factors most commonly reported are male sex and tobacco use. Only 30% of patients with SISMAD are hypertensive, while hypertension plays a much more important role in combined aortic and superior mesenteric artery dissection [3].

The etiology of SISMAD needs to be better established. Even though a few conditions include muscular dysplasia, medial cystic necrosis, arterial mediolysis, adventitial inflammation, disruption of the internal elastic lamina, penetrating arterial ulcer, pseudoaneurysm, and aneurysms were reported to be associated with SISMAD, no evidence supports any specific pathologies [4,5]. SISMAD might be familial, as evidenced by a report on a specific genetic locus previously linked to familial ascending aortic aneurysms and dissections [5]. A histological examination of the patient sample showed myxoid degeneration of the SMA wall [6]. The dissection entry was reported to be within 1.5-6.0 cm from the origin of SMA [7], hence another hypothesis for SISMAD etiology has been postulated that SISMAD might result from shear stress affecting the area of convexity of SMA [3].

Clinical presentation of SISMAD varies, but most patients present with abdominal pain, as with our patient who complained of periumbilical pain. Other symptoms reported range from abdominal distention, nausea, vomiting, diarrhea, melena, and postprandial pain, which can last for months, to more severe presentations such as intestinal obstruction or shock [4,8]. Sometimes, SISMAD is found accidentally on CT scans and autopsy [4]. Several CT-based classifications have been proposed [9], but no substantial evidence supports the correlation between SISMAD subtypes and clinical outcomes, thereby explaining the lack of consensus on which classification should be used.

To relieve symptoms and prevent complications are the aims of treatment for SISMAD. Surgery is the final option, reserved for patients showing signs of bowel infarction or SMA rupture. The choice between conservative versus endovascular stenting, is somewhat more controversial [2].

The indication for endovascular stenting is not clear, but it is usually chosen when the abdominal symptoms are prolonged. Patients undergoing endovascular stenting have been reported to have a lower recurrence of abdominal pain [8].

Abdominal pain can be relieved without the need of stenting or even bowel rest. Intravenous heparin could be used for 5 days [5], whereas 5-7 days is standard practice at our institution, while antithrombotic therapy is not recommended [10]. Sixty-two patients were included in an observational study by Shi et al, in which 16 patients were considered

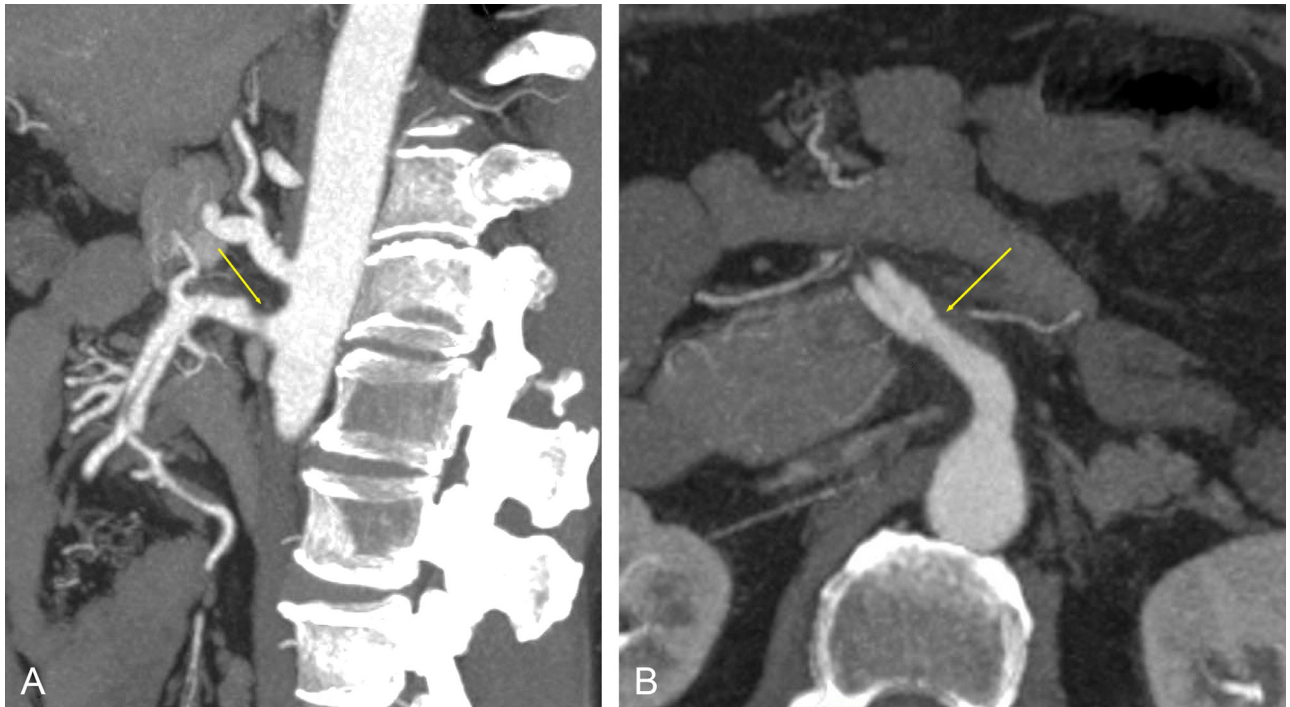


Fig. 1 - (A) Intimal flap in the superior mesenteric artery; (B) partially thrombosed false lumen (yellow arrow).

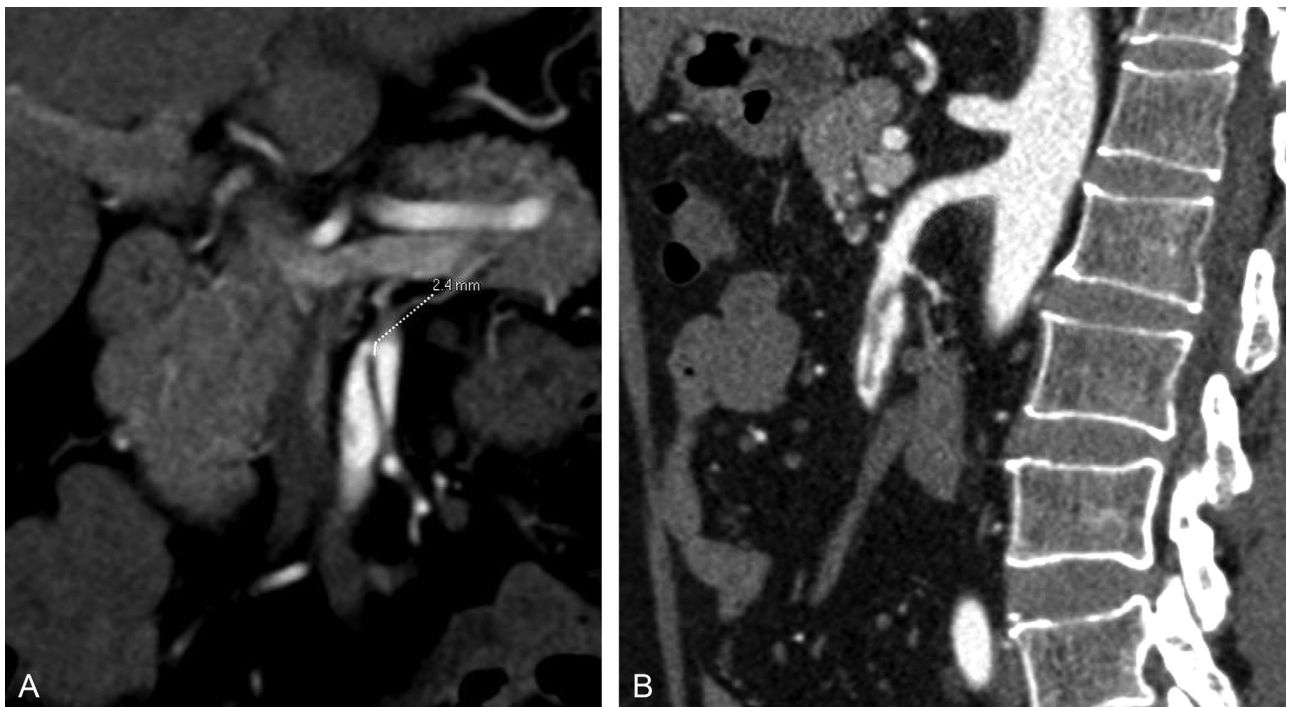


Fig. 2 - Computed Tomography at 6 months after discharge: (A) the diameter of the entry point was 2.4 mm, (b) SMA viewed sagittally.

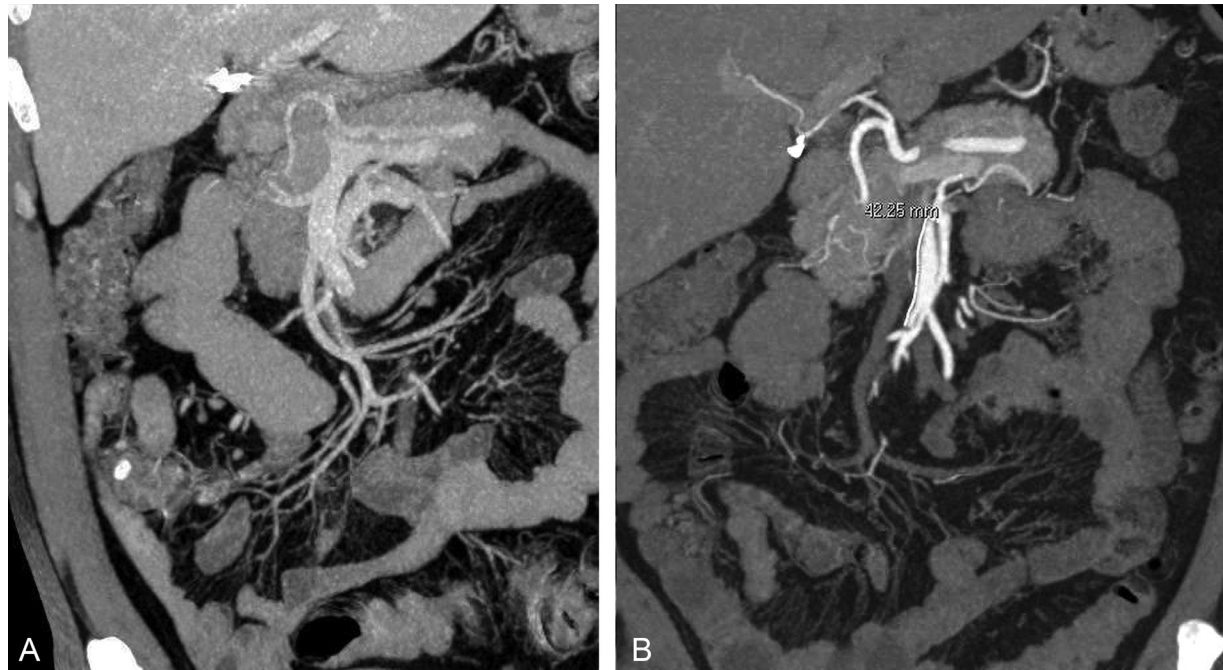


Fig. 3 – Before (A) and after 6 months (B): no signs of bowel ischaemia. The distal branches of the superior mesenteric artery were patent.

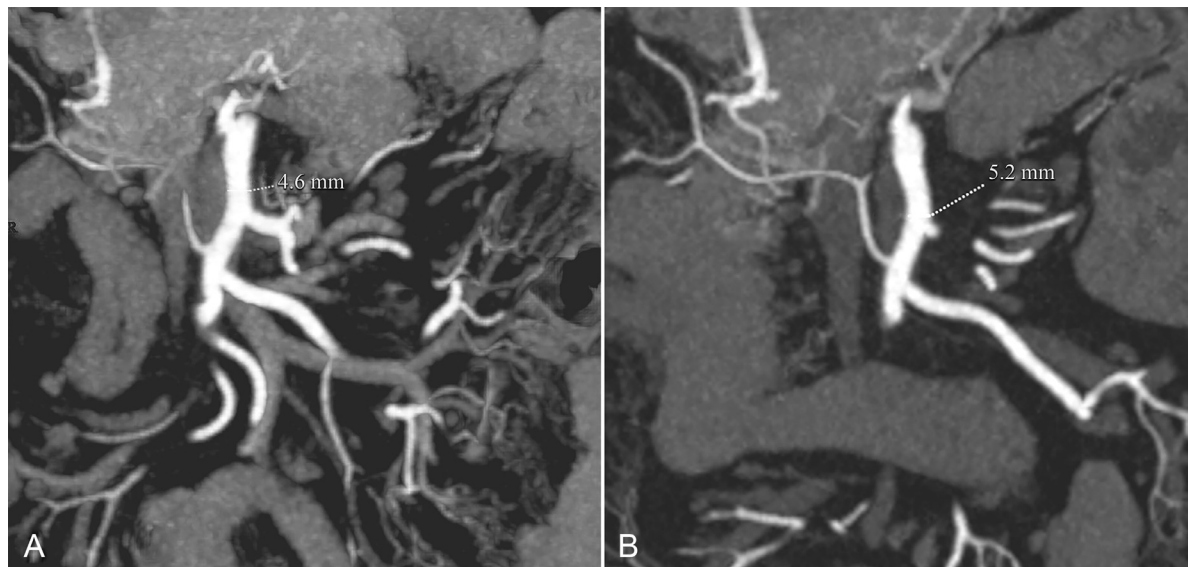


Fig. 4 – Computed tomography scans post-discharge (A) at 1 year, the diameter of true lumen was 4.6 mm and (B) at 1.5 year, the diameter of true lumen was 5.2 mm. At both points, the distal branches of the superior mesenteric artery were patent.

high risk. The authors suggest a dissection longer than 5 cm is a predictor of patients who may need stenting or surgery, and conservative management could be applied for most low risk patients [7]. In our case, the dissection length was 4.2 cm, and the patient was treated conservatively with long-term anticoagulant and regular follow-ups. His condition showed improvement in terms of symptomatology and radiology. This, once again, might suggest that not every patient must be considered for endovascular stenting and, there-

fore, can avoid the iatrogenic complications of more invasive interventions.

Conclusion

SISMAD should be considered as part of differential diagnoses when patients in their fifth to seventh decades of life

present with acute abdominal pain. Treatment includes conservative management, percutaneous endovascular interventions, or surgery, but most patients can be managed conservatively.

Patient consent

Written, informed consent was obtained from this patient about the case report that we are submitting for publication.

Declarations

Ethical approval and consent to participate

All procedures performed in studies involving the human participant were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent was obtained from the individual included in the study.

Consent for publication

Consent for publication was obtained for the individual's data included in the study.

Availability of data and material

The datasets generated and/or analysed during the current study are not publicly available in order to protect patient privacy but are available from the corresponding author on reasonable request.

Authors' contribution

NMV, VDAB and DTN collected the necessary data. NMV and DN was responsible for writing. DV and NJ reviewed the case report. THN and LMBT provided scientific supervision.

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