

Case Study

Visceral artery aneurysms: A complex and unresolved issue

ABSTRACT

Aims: The lack of prospective studies on abdominal non-aortic true or false aneurysms results in insufficient data to predict their natural history and propose a treatment of choice. The experience provided by case reports is important to discuss their management and prevention. This is **relevant** because abnormal enlargements in superior mesenteric artery are relatively uncommon but associated with specific symptoms and a high incidence of rupture and/or ischemic bowel complications

Presentation of Case: Open surgery was preferred in a giant true aneurysm with an associated arteriovenous fistula, due to the likely need for revascularization and probable lack of stability for a stent graft. Contrarily in a pseudoaneurysm with a similar size we performed coil embolization. Incidentally, this procedure required a subsequent deployment of a covered stent graft to resolve bleeding.

Discussion: We discuss the outcomes of true and false aneurysms in the superior mesenteric artery, which required urgent treatment due to their high diameter. Their extremely large size also suggests a protracted course and a considerable period of time without proper surveillance through imaging techniques. Endovascular techniques seem to be favored in the elective setting and open surgery in the emergent setting but the challenge for the vascular surgeon to choose a treatment is considerable. Patient's clinical state, perceived risk of rupture, the size of the lesion and expected quality of life conditioned the choice.

Conclusion: The described cases illustrate the urgent requirement of treatment guidelines and/or screening programs to detect abdominal non-aortic aneurysms at an early stage.

Keywords: Visceral artery aneurysms; pseudoaneurysms; risk assessment; screening; superior mesenteric artery

1. INTRODUCTION

Aneurysms involving splenic, gastro-duodenal, hepatic, superior mesenteric and renal arteries are commonly referred as visceral artery aneurysms (VAAs). Pseudoaneurysms (VAPAs) are contained ruptures in the respective vessel wall with collection of blood. Both may result in massive hemorrhage. Their actual prevalence is difficult to calculate because the use of rapid and relatively simple imaging techniques is associated with an apparently growing incidence.

It is generally accepted that true aneurysms may be unrelated to the presenting complaint, whereas pseudoaneurysms are most likely symptomatic. This assumption is apparently inaccurate in superior mesenteric artery (SMA) enlargements, which are commonly associated with abdominal pain and a high operative mortality rate. Case reports are the only sources of available data to characterize this condition and, consequently, there are no clinical guidelines [1-5]. However, current clinical experience increasingly suggests that

endovascular repair in most cases represent a promising alternative to the more classical open-surgery procedures. Here we describe two observations illustrating the challenge that represent the different diagnostic procedures, assessment of risk and therapeutic alternatives. Conservative options (i.e., watchful waiting) were not considered due to the size and risk of rupture and the Surgical Review Board of the Hospital Joan XXIII has previously approved the possible interventional procedures. Informed consents were obtained according to guidelines provided by the Ethics Committee (OBESPAD 14-07-31/7proj3).

2. PRESENTATION OF CASES

2.1 Case 1

A 68 year-old, non-smoker female was admitted to our Hospital presenting an 8-hour history of intense and oppressive thoracic and abdominal pain, which radiated to the neck and was accompanied by diaphoresis and vomiting. Myocardial infarction was discarded and a large, firm and pulsatile mass was palpable in the abdomen with a clear murmur under auscultation. She had a past medical history of rheumatic fever, which required the implant of a mechanical prosthetic mitral valve and left hemiplegia resulting from a previous stroke during an episode of postpartum infective endocarditis. A computed tomography (CT) scan showed an 11-cm (in diameter) true aneurysm at the distal segment of SMA, the concomitant presence of peripheral calcification and an arteriovenous fistula causing enlargement of the mesenteric vein (Figure 1, A-C). A medial laparotomy was performed and the adhesions to the adjacent tissue were removed to control the proximal and distal aneurysm necks. Other vessels present in the anatomical area of influence of the aneurysm were provisionally clamped (more than 30 min) without apparent signs of ischemia (Figure 1, D). Thus, the aneurysmal sac was opened, the retrograde bleeding from the fistula contained, and full resection of aneurysmal sac was performed without grafting. The abdomen incision was then maintained open under a negative pressure abdominal closure dressing and the patient was carefully monitored at the Intensive Care Unit (ICU). Approximately 24 hours after this surgical procedure there was an acute severe ischemic event affecting portions of jejunum and ileum that required surgical intervention. The patient died 24 hours after this further intervention due to related complications. The search in blood and tissues for infective agents was negative and the histology in portions of the excised artery depicted advanced atherosclerosis.

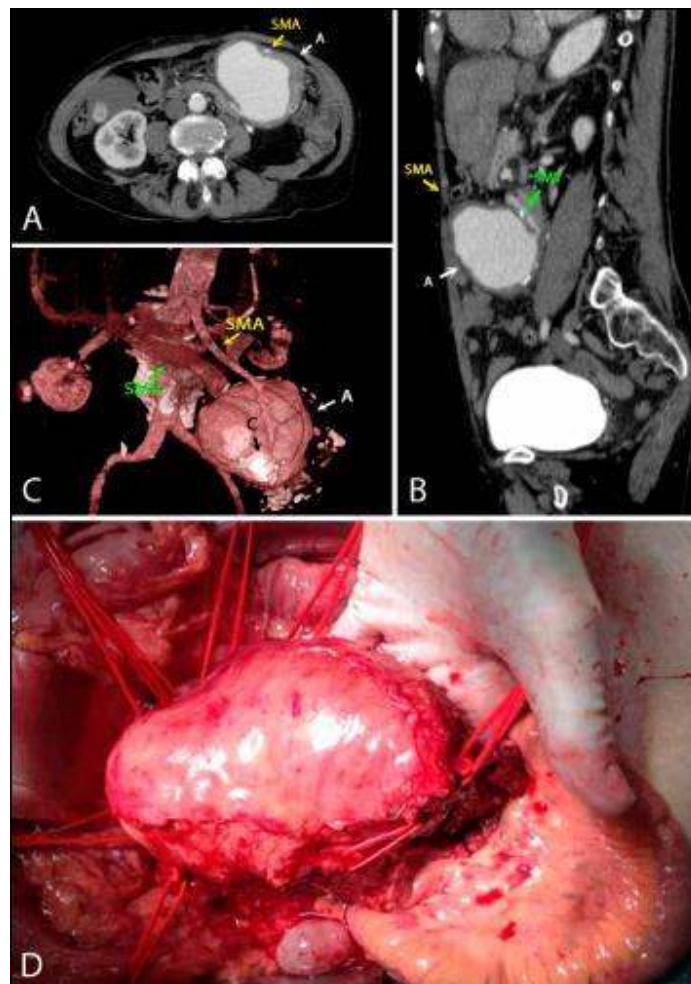
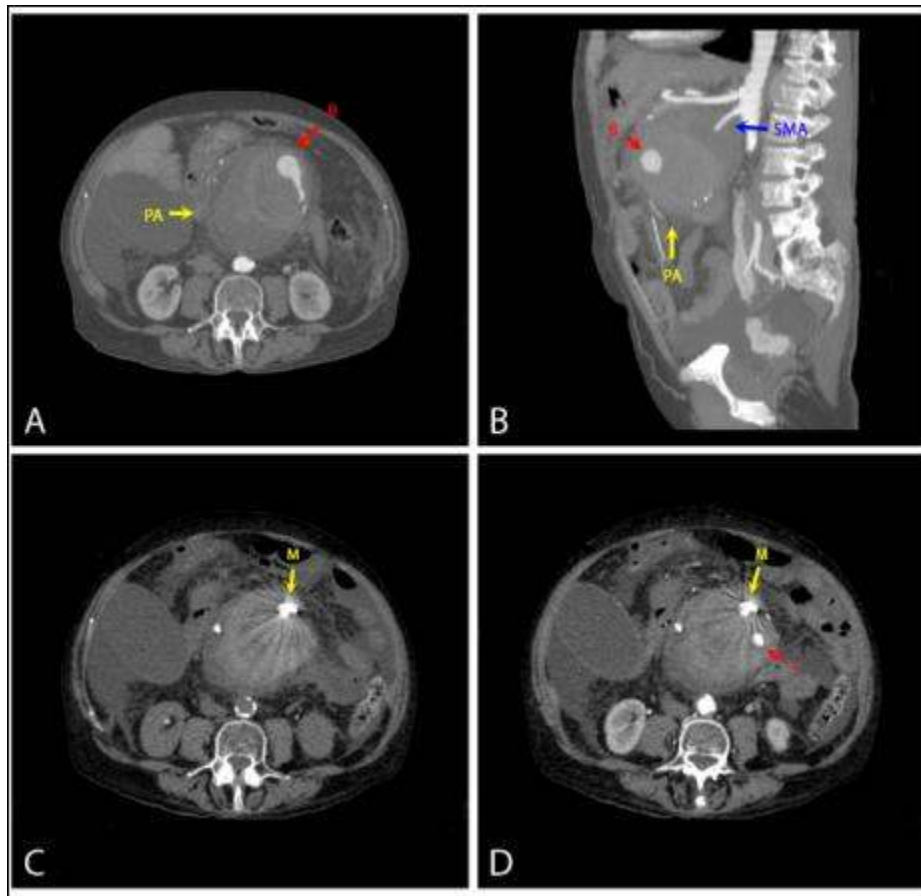


Fig. 1. Management of visceral artery aneurysms. Non-invasive imaging techniques, in particular computed tomography angiogram scans were important for the diagnosis of a giant true aneurysm (A) arising from the distal branches of the superior mesenteric artery (SMA) and an arteriovenous fistula causing an enlargement of the superior mesenteric vein (SMV) (A,B). Other anatomical details were provided by a three-dimensional reconstruction, including the present of calcification (C). Clamping of the aneurysmal sac for more than 30 min apparently excluded the presence of ischemic bowel (D), a procedure that should be re-assessed according to the unsatisfactory outcome.

2.2 Case 2

A 58-year-old female with a history of smoking and high alcohol intake was admitted to our Hospital presenting a 6-hour intense and diffuse abdominal pain, nausea and vomiting. Clinical records depicted a past medical history of chronic pancreatitis, liver cirrhosis, portal hypertension, and non-bleeding esophageal varices. A palpable, painful and pulsatile mass was found in epigastrium. Laboratory studies and subsequent arteriography and CT scans suggested the provisional diagnosis of an arterial pseudoaneurysm, which was complicating active pancreatitis. We found a 10-cm (in diameter) blood collection with signs of a recent bleeding involving SMA (Figure 2, A, B). In this case, the decision was to perform endovascular treatment. The brachial artery was punctured and the SMA was catheterized

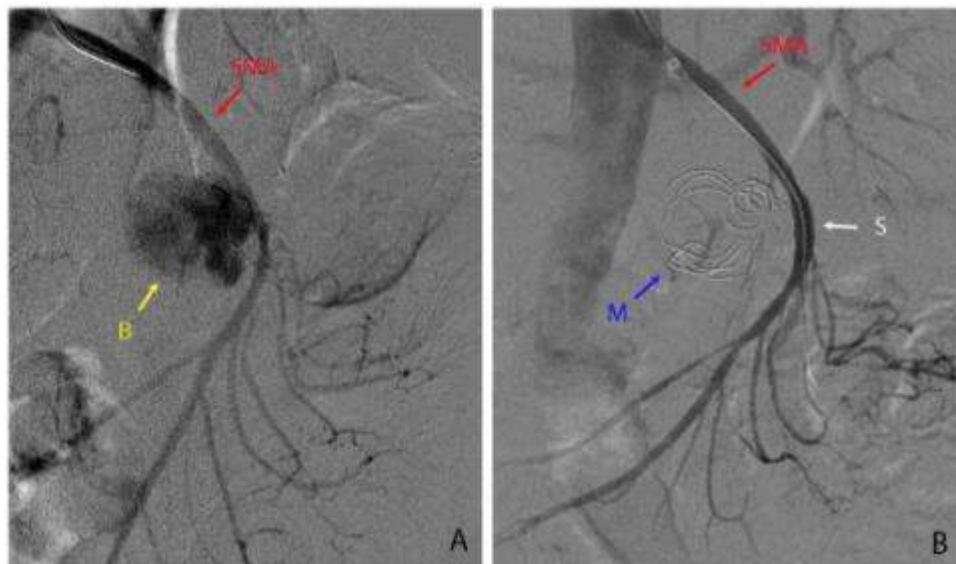
82 to reach the pseudoaneurysm neck with a Progreat™ microcatheter (Terumo, Tokyo,
83 Japan). We first released hilal embolization microcoils™ (Cook Medical, Bloomington, IN,
84 USA) at the bleeding point but control CT scans depicted a leak providing a persistent inflow
85 filling of the pseudoaneurysmatic sac through the microcoils (Figure 2, C, D). Then, a
86 covered stent graft (Bentley InnoMed, Hechingen, Germany) was positioned. The complete
87 resolution of the endoleak and the patency of the stent graft were verified through
88 angiography (Figure 3). After 3 days of follow-up at the ICU, the patient was referred to their
89 attending physicians at the Gastroenterology Department without further related vascular
90 complications.



91
92 **Fig. 2. A giant pseudoaneurysm illustrates the value of endovascular treatment in**
93 **hostile environment for abdominal surgery.** Contrast-enhanced computed tomography
94 scans before endovascular treatment (A, B) depicted a superior mesenteric
95 pseudoaneurysm (PA) with signs of active bleeding (B). Further imaging studies after
96 endovascular management confirmed that microcoils were into the aneurysm sac (C) but
97 also the persistence of a leak (L) indicating an unresolved blood inflow (D), which required a
98 stent graft.
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Fig. 3. Arteriography remains a valuable tool in surgical management. Findings in Figure 2 (C, D), including active bleeding (B) were further confirmed by angiography demonstrating the value of this technique for further diagnosis (A). The procedure also detected the presence of microcoils (M) and guided the deployment of the covered stent in the mesenteric superior artery (SMA), which resolved this clinical condition (B).

115 3. DISCUSSION

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Patients with VAAs have intact arterial layers but in VAPAs the arterial wall is incomplete. The described cases illustrate the need to collect epidemiological data on the prevalence, incidence and mortality rate of abdominal non-aortic true or false aneurysms (i.e., abnormally enlarged visceral arteries). The reported prevalence is not different from that estimated for abdominal aortic aneurysms (AAA) (approximately 2%) and their threatening complications are potentially avoidable [6]. The frequency of occurrence of VAAs and VAPAs is apparently increasing as a result of incidental discoveries during imaging examinations and many true aneurysms and essentially all pseudoaneurysms require treatment [7,8]. The size of lesions in our two case reports in combination with the perceived risk for the patient indicated urgent treatment because complications were likely and life threatening (e.g., thrombosis and/or embolization, compression of adjacent structures and hemorrhage from rupture). Aneurysms in SMA represent a 5-7% of VAAs but mortality is apparently higher than that observed in other arteries, especially when the treatment is not elective [9]. Trauma, infection and even degenerative processes have been formerly described among the causes of aneurysms but atherosclerosis is now recognized as the primary event in the pathogenesis of these disorders [4,5,9]. Consequently, it is important to reinforce the strategy of risk-factor modification, which might reduce the rate of aneurysmal growth (i.e., cessation of smoking, treatment of hypertension and/or of hypercholesterolemia, or drugs that inhibit the renin-angiotensin pathway and/or the expression of metalloproteinases) [10].

137 Unlike other VAAs, SMA aneurysms (true and false) are symptomatic. However, laboratory
138 data, symptoms and signs are not sufficient to discriminate between true and false
139 aneurysms and require the use of imaging techniques. There are no gender differences in
140 distribution but the incidence of ischemic bowel and rupture is apparently high [8,11]. Clinical
141 manifestations include abdominal pain and a palpable and pulsatile abdominal mass [6].
142 This fact is clearly illustrated in the current cases report and it suggests the importance
143 of contrast material-enhanced abdominal CT examination for rapid diagnosis. Magnetic
144 resonance imaging represents a valid alternative because the new techniques provide
145 additional vascular information. Ultrasonography was useless in the management of these
146 patients but angiography remained as an important tool for treatment and further diagnosis.

147 The choice of treatment is a challenging task. The clinical condition of the two patients in the
148 current study was not comparable but size and shape was determinant to guide decisions. In
149 both cases the Laplace's law (i.e., Tension = Pressure x Radius) suggests a high rate of
150 expansion and even the adoption of a spherical shape could not limit the expansion due to
151 the increased wall tension. Some authors consider open-surgery a better choice for VAAs
152 and VAPAs of this size, but the current experience in endovascular treatment is apparently
153 contradictory to this assumption and a matter of discussion [12-17]. Similar clinical situations
154 will require a prompt response and decision from the vascular surgeon. Open-surgery may
155 provide definitive treatment (i.e., higher durability and reduced need for follow-up studies),
156 which includes the possibility for a better evaluation of the need for revascularization of the
157 affected organs. This is especially important in arteries such as SMA with inadequate flow or
158 others without collateral flow. On the other hand, catheter-based interventions may be
159 difficult to perform in the emergency setting and anatomy, size and predicted complications
160 should be carefully considered. Hence, the goal should be to develop ways for exclusion of
161 the aneurysm's sac from the circulation and to preserve distal flow. Although most deaths
162 occur in patients undergoing urgent repair [1,5,15,18], elective treatments (after surveillance
163 and/or medical treatment) were considered unrealistic in these cases.

164 In case 1, we chose open surgery due to the perceived high risk of rupture, the likely need
165 for revascularization and a probable lack of stability for a stent graft [4]. We also predicted
166 the likelihood of multiple vessels originating from the proximal and distal necks of the
167 aneurysm and that endovascular treatment will probably result in a conversion to an open
168 surgery procedure (i.e., over-imposed complications). Because clamping for more than 30
169 minutes did not depict intestinal ischemic changes, the procedure was considered
170 successful but this decision requires further evaluation because ischemic events and
171 subsequent intestinal resection are not uncommon. In case 2, however, the performed
172 laboratory studies suggested the presence of acute pancreatitis (namely increased serum
173 levels of amylase and lipase). These results combined with those provided by imaging
174 techniques clearly suggested the presence of an hostile environment for abdominal surgery
175 and we decided to perform coil embolization as a minimally invasive endovascular
176 technique, which provides a decreased length of hospital stay, and better quality of life. Of
177 note, a leak was further resolved by the deployment of a covered stent graft [19,20]. This
178 procedure occluded two arterial branches but without clinical signs of ischemia and a
179 satisfactory follow-up. Knowledge and tools designed to improve endovascular treatment are
180 in constant expansion. Although some complications have been reported, current
181 alternatives include the use of flow-diverting stents or multilayer fluid modulating stents
182 designed to reduce the flow velocity allowing for physiological thrombosis [13,17]. Our
183 experience suggests that open surgery should be the first choice for unstable patients with
184 established rupture or in high clinical risk but endovascular repair, once limited to elective
185 options and stable patients, deserves further consideration in more patients.

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4. CONCLUSION

The frequency of occurrence of VAAs/VAPAs is considerable. SMA aneurysms are relatively uncommon but frequently symptomatic and represent a dangerous type of VAA because of the high incidence of ischemic bowel complications. All SMA pseudoaneurysms are symptomatic and should be suspected in the presence of concurrent inflammatory diseases (especially pancreatitis). There are no prospective studies to properly define their prevalence and natural history. Therefore, the indication of treatment remains individualized and likely determined by the size and the perceived risk of rupture. Current approach for VAAs/VAPAs is early intervention and endovascular techniques seem to be favored. These are especially considered in the elective setting but open surgery is likely the preferred method in giant aneurysms in the emergent setting. Our data illustrate that perhaps it is time to seriously consider screening policies to detect minor lesions and an active approach to follow-up their likely expansion.

CONSENT

Written informed consent was obtained from one patient and the next of kin from other for publication of details of these case reports, including images, ensuring anonymity for the eventual readers. Copies are available for review by the Editorial Office/Chief Editor/Editorial Board members of this journal.

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273	DEFINITIONS, ACRONYMS, ABBREVIATIONS
274	AAA: Abdominal aortic aneurysm
275	CT: Computed tomography
276	ICU: Intensive care unit
277	SMA: Superior mesenteric artery
278	VAA: Visceral artery aneurysms
279	VAPA: Pseudoaneurysms