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 important 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: These cases illustrate that treatment guidelines are urgently required and the need for 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 (VAA). Pseudoaneurysms (VAPA) are contained ruptures of the respective vessel wall. Both may result in massive hemorrhage. Their actual prevalence is difficult to calculate 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 source 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 possible interventional procedures have been previously approved by the Surgical Review Board of the Hospital Joan XXIII. Informed consents were obtained according to guidelines provided by the Ethics Committee (OBESPAD 14-07-31/7proj3).

2. PRESENTATION OF CASE

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 and 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). In view of that, 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 procedure 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.

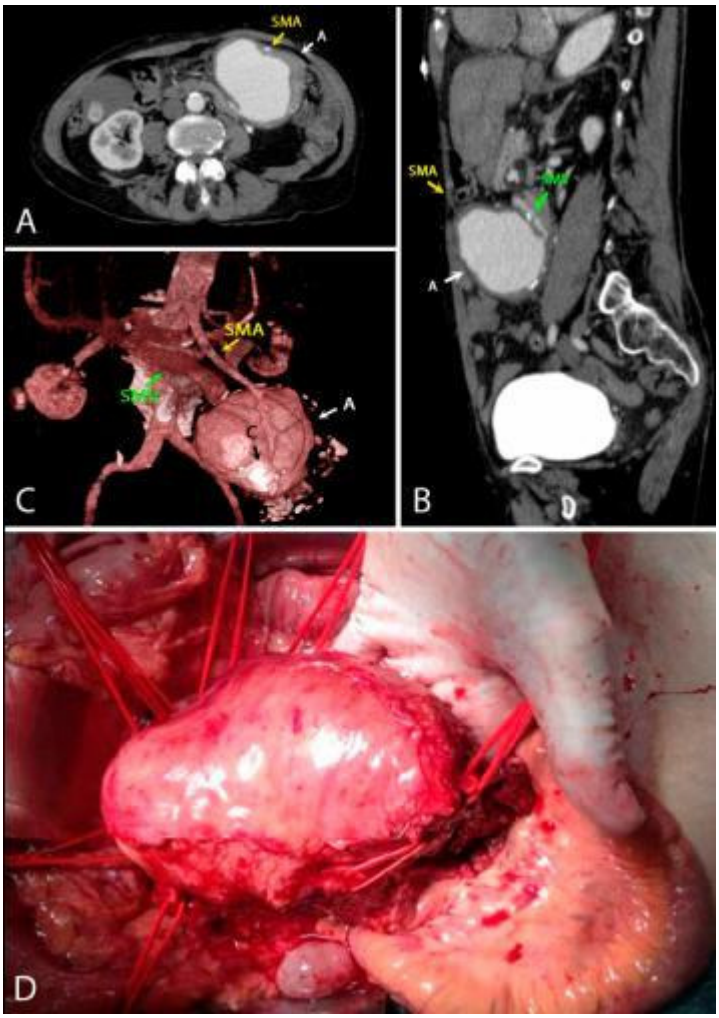

Fig. 1. Management of visceral artery aneurysms. Non-invasive imaging techniques, in particular computed tomography angiogram scans were important for the diagnosis of a giant true aneurysm (A) arising from the distal branches of the superior mesenteric artery (SMA) and an arteriovenous fistula causing an enlargement of the superior mesenteric vein (SMV) (**A,B**). Further 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 ulterior 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, the presence of 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 confirmed our provisional diagnosis of arterial pseudoaneurysm complicating 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 to reach the pseudoaneurysm neck with a Progreat™ microcatheter (Terumo, Tokyo, Japan). We first

released hilal embolization microcoils™ (Cook Medical, Bloomington, IN, USA) at the bleeding point but control CT scans depicted a leak providing a persistent inflow filling of the pseudoaneurysmatic sac through the microcoils (Figure 2, C, D), and a covered stent graft (Bentley InnoMed, Hechingen, Germany) was positioned. The complete resolution of the endoleak and the patency of the stent graft were verified through angiography (Figure 3). After 3 days of follow-up at the ICU, the patient was referred to their attending physicians at the Gastroenterology Department without further related vascular complications.

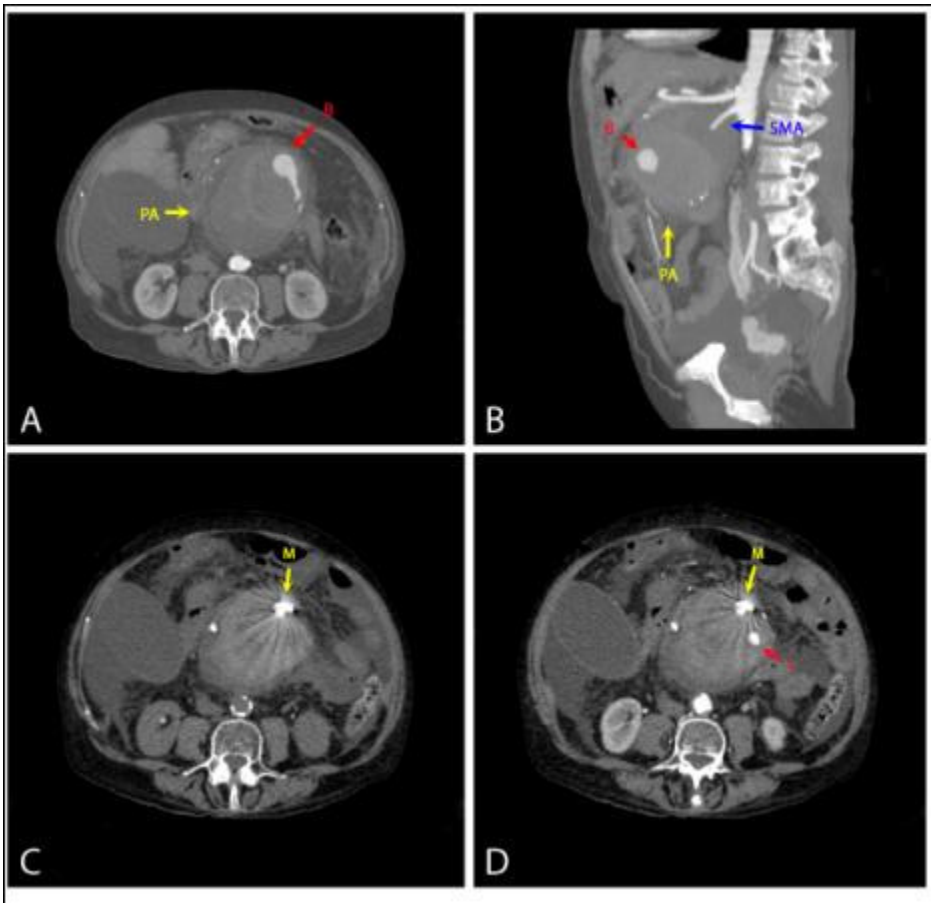

Fig. 2. A giant pseudoaneurysm illustrates the value of endovascular treatment in hostile environment for abdominal surgery. Contrast-enhanced computed tomography scans before endovascular treatment **(A, B)** depicted a superior mesenteric pseudoaneurysm (PA) with signs of active bleeding (B). Further imaging studies after endovascular management confirmed that microcoils were into the aneurysm sac **(C)** but a leak (L), a continuous unresolved blood inflow persisted **(D)**.

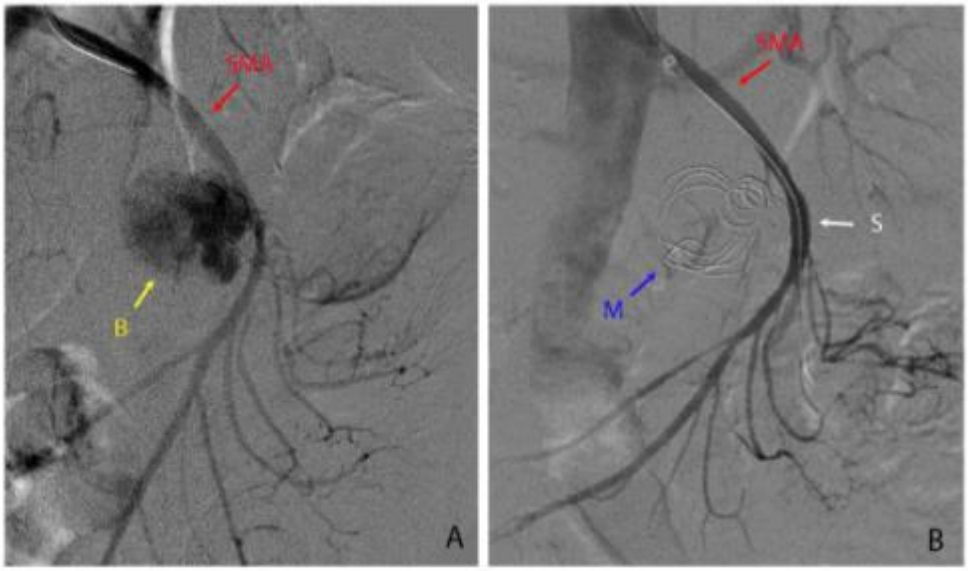

Fig. 3. Arteriography remains a valuable tool in surgical management. Findings in Figure 2 (C, D), including active bleeding (B) were further confirmed by angiography demonstrating the value of this technique for further diagnosis **(A)** and treatment to detect the presence of microcoils (M) and to guide the deployment of the covered stent in the mesenteric superior artery (SMA), which resolved this clinical condition (**B)**.

3. DISCUSSION

Patients with VAA have intact arterial layers but in VAPA the arterial wall is incomplete. For the above-mentioned reasons, there is an obvious 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%) [6] and their threatening complications are potentially avoidable. The frequency of occurrence of VAA and VAPA 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 VAA but mortality is apparently higher than that observed in other arteries, in particular when treatment is not elective [9]. Trauma, infection and even degenerative processes have been formerly described as the causes of this disease but atherosclerosis is now recognized as the primary event in pathogenesis [4,5,9]. This is important because the strategy of risk-factor modification might reduce the rate of aneurysmal growth (e.g., cessation of smoking, treatment of hypertension and/or hypercholesterolemia, or drugs that inhibit the renin-angiotensin pathway and/or the metalloproteinases) [10].

Unlike other VAA, SMA aneurysms (true and false) are symptomatic. There are no gender differences in distribution but the incidence of ischemic bowel and rupture is apparently higher [8,11]. Clinical manifestations include abdominal pain and a palpable and pulsatile abdominal mass [6]. This fact is clearly illustrated in our cases reports and suggests the value of contrast material-enhanced abdominal CT examination for rapid diagnosis.

Magnetic resonance imaging was also an alternative because new techniques provide valuable vascular information. Ultrasonography was useless in the management of these patients but angiography was important during treatment and for further diagnosis.

The choice of treatment is a challenging task. The clinical condition of these two patients was not comparable but size and shape was determinant to guide decisions. In both cases the Laplace's law (i.e., Tension = Pressure x Radius) suggests a high rate of expansion and even the adoption of a spherical shape could not limit expansion and exceeding wall tension. Some authors consider open-surgery a better choice for VAA and VAPA of this size, but current experience in endovascular treatment is apparently contrary to this assumption and a matter of dispute [12-17]. The described clinical situations require a prompt response and decision from the vascular surgeon. Open-surgery may provide definitive treatment (i.e., durability and reduced need for follow-up studies), which includes the possibility to better evaluate the need for revascularization of affected organs. This is especially important in arteries such as SMA with inadequate flow or others without collateral flow. On the other hand, catheter-based interventions may be difficult to perform in the emergency setting and anatomy, size and predicted complications should be carefully considered. The goal should be the exclusion of the aneurysmal sac from the circulation and to preserve distal flow. Although most deaths occur in patients undergoing urgent repair [1,5,15,18], elective treatments (after surveillance and/or medical treatment) were considered unrealistic in these cases.

In case 1, we chose open surgery due to the perceived high risk of rupture, the likely need for revascularization and a probable lack of stability for a stent graft [4]. We also predicted the likelihood of multiple vessels originating from the proximal and distal necks of the aneurysm and that endovascular treatment will probably result in a conversion to an open surgery procedure (i.e., over-imposed complications). Because clamping for more than 30 minutes did not depict intestinal ischemic changes, the procedure was considered successful but this decision requires further evaluation because ischemic events requiring intestinal resection is not uncommon. In case 2, however, laboratory studies suggested the presence of acute pancreatitis (i.e., an expected hostile environment for abdominal surgery) and we decided to perform coil embolization as a minimally invasive endovascular technique, which provides a decreased length of hospital stay, and better quality of life. Imaging techniques, however, revealed the presence of a leak that was further resolved by the deployment of a covered stent graft [19,20]. This procedure occluded two arterial branches but without clinical signs of ischemia and a satisfactory follow-up. Knowledge and tools designed to improve endovascular treatment are in constant expansion. Current alternatives include the use of flow-diverting stents or multilayer fluid modulating stents designed to reduce the flow velocity allowing for physiological thrombosis but complications have also been reported [13,17]. Our experience suggests that open surgery should be the first choice for unstable patients with established rupture or in high clinical risk but endovascular repair, once limited to elective options and stable patients may be extended to most patients.

4. CONCLUSION

The frequency of occurrence of VAA/VAPA 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 inflammation (especially from 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 VAA/VAPA 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 the emergent setting and in the presence of giant aneurysms. Our data illustrate that perhaps it is time to seriously consider screening policies to detect minor lesions and programs to follow-up their likely expansion.

CONSENT (WHERE EVER APPLICABLE)

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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DEFINITIONS, ACRONYMS, ABBREVIATIONS

AAA: Abdominal aortic aneurysm
CT: Computed tomography
ICU: Intensive care unit
SMA: Superior mesenteric artery
VAA: Visceral artery aneurysms
VAPA: Pseudoaneurysms