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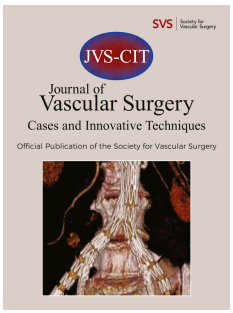

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Presentation and Management of Rare Saccular Superior Mesenteric Artery Trunk and Branch Aneurysms

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ARTICLE HIGHLIGHTS

Type of Research: Single-center retrospective cohort study

Key Findings: Open aneurysmectomy of 5 cases of superior mesenteric artery aneurysms yielded favorable outcome without need for bowel resection in 4 patients without rupture, but mortality in 1 case of rupture.

Take home Message: Open aneurysmectomy is a feasible treatment option for cases of superior mesenteric artery aneurysms.

Abstract

Superior mesenteric artery (SMA) aneurysm (SMAA) is caused by degeneration of the visceral arteries. Although a rarely encountered entity, it requires timely management due to the high mortality rate associated with rupture, particularly when the aneurysm is saccular in nature. As such, urgent treatment is generally indicated. We present 5 cases of SMAA arising from the main trunk or branches of the SMA.

Keywords: superior mesenteric artery, aneurysm, rupture, revascularization

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Introduction

Visceral artery aneurysms are rarely encountered, with an incidence of 0.1%-2%. Of this group, superior mesenteric artery (SMA) aneurysms (SMAA) account for just 5.5%, third in line behind splenic and hepatic artery aneurysms[1]. Most SMAAs present as incidental findings, however some patients have reported a combination of nausea, weight loss, malaise, pyrexia, and abdominal pain[2]. A combination of a low index of suspicion, stemming from rarity of the

disease, and non-specific symptoms usually leads to a delay in diagnosis, which can have lethal consequences as the reported mortality for a ruptured SMAA is 38%-50%[3]. Since a subset of SMAAs are mycotic in origin we expect clinicians to increasingly encounter SMAAs with the rise in intravenous drug abuse (IVDA) in relation to infective endocarditis[4]. Therapy is mainly divided into open surgical approaches including aneurysmectomy with or without vascular reconstruction although success with endovascular techniques has been reported, with long-term antibiotic therapy in cases of mycotic aneurysms. In all scenarios, end-organ resection is considered if perfusion to the small or large intestines is thought to be jeopardized[5]. Only recently, the Society of Vascular Surgery released guidelines pertaining to the diagnosis, management, and follow-up of visceral arterial aneurysms, including SMAA[6]. We aim to present a single institution's experience with aneurysms of the SMA or its branches. All patients consented for publication.

Methods

Medical records and imaging studies from a single academic medical center from January 2016 to January 2019 were retrospectively reviewed. We describe the presentation, operative management, post-operative outcomes, and follow up from a series of five patients with aneurysms; 4 originating from a branch of the SMA, and the 5th from the main trunk of theSMA.(**Table 1 and 2).**

Case Reports

Case 1

A 75-year-old woman with lower back pain underwent computed tomography (CT) with intravenous (IV) contrast to evaluate for lumbar spine disease. She had no history of IVDA,

endocarditis, degenerative disease, aortic or visceral aneurysms, or peripheral arterial disease. An incidental finding of a 2.6 x 3.5 cm saccular aneurysm of the SMA arising from a proximal branch of the SMA was noted **(Figure 1).** The patient was taken electively for laparotomy. On exploration it was found that the SMAA was compressing the celiac artery, which appeared to be chronically occluded. Because the SMA was providing significant collateral flow to the hepatic and splenic territories as revealed on angiography 1 month prior, we opted to preserve flow with vascular reconstruction. The aneurysm was significantly adherent to surround tissue including the duodenum, and careful mobilization of the duodenum away from the aneurysm was performed. She underwent aneurysmectomy, and, as clinical suspicion for an infectious etiology was low, an 8-mm prosthetic polytetrafluoroethylene graft was used to create an aorto-common hepatic artery bypass. Approximately 20-30 minutes after the anastomosis, the bowel was reassessed and found to have a pink color, peristalsis, and a palpable pulse and Doppler signal at the mesenteric border. The patient's recovery was uncomplicated. Intraoperative cultures were negative. Two weeks post-operatively, a duplex mesenteric study showed a patent aorto-hepatic artery bypass. She continues to follow up in our clinic two years later, with further duplex scans showing both bypass and SMA patency.

Case 2

A 54-year-old woman presented with lower back pain and underwent a CT scan of the lumbar spine with IV contrast and was found to have a 1.5 cm aneurysm arising 4 cm distal to the take-off of the SMA. She had no risk factors for SMA aneurysms in her history. Of note, she did undergo a laparoscopic tubal ligation 13 years prior. Although speculative, an injury at that time could have resulted in visceral arterial degeneration and aneurysm formation. The aneurysm had a worrisome configuration with a saccular component. While the aneurysm involved the

main trunk, two major branches of the SMA originated directly from the aneurysm. Reconstruction was performed with a bifurcated vein bypass graft using reversed great saphenous vein **(Figure 2).** Intraoperative cultures were negative, yielding a diagnosis of cryptogenic SMAA. Post-operative surveillance CT angiography (CTA) showed patent vein bypass graft at 30 days, and she reported resolution of her lower back pain on follow-up.

Case 3

A 74-year-old female was admitted with right upper quadrant abdominal pain and underwent a CT angiogram of the abdomen and pelvis. A 1.2 cm saccular aneurysm at the branchpoint of the SMA and ileocolic artery was identified **(Figure 3)**. She had a history significant for *Staphylococcus epidermidis* mitral valve endocarditis treated with open mitral valve repair less than a month prior to presentation. Blood cultures at time of admission were positive for *S. epidermidis*. She admitted to recent weight loss and decreased appetite, though this had been previously attributed to the post-operative recovery following her mitral valve repair. On laparotomy, an aneurysmal ileal branch off of the SMA was easily identified with cephalad retraction of the small intestines. She underwent aneurysmectomy, isolating the ileal branch proximally and distally, while ligating feeding vessels. Intraoperative cultures grew *S. epidermidis*. The acute care surgery was present for intraoperative observation at initial laparotomy and again prior to closure of the abdomen and based on the appearance of the bowel as well as its rapid peristalsis, determined there was no need for bowel resection nor second-look laparotomy. The patient recovered and was discharged on a 6-week course of daptomycin.

Case 4

A 70-year-old male presented to the emergency department complaining of left-sided chest pain. He had a history of IVDA, cocaine abuse, and multiple episodes of genital chlamydia

and gonorrhea infections over the past year. At the time of presentation, the patient had no identifiable infection nor leukocytosis, and had negative blood cultures. CTA of the chest and abdomen showed penetrating aortic ulcers of the descending thoracic aorta without rupture as well as an incidental, 2 cm saccular aneurysm of the SMA approximately 6 cm from the origin of the SMA. Aneurysmectomy was performed without violating the main SMA trunk. Revascularization and end-organ resection were not required. The aneurysm was opened off the surgical field and yielded purulent fluid. Post-operatively, cultures of this fluid did not isolate microorganisms. The patient was treated with IV antibiotics for 4 weeks, then underwent thoracic endovascular aortic repair to treat the penetrating ulcers of his descending thoracic aorta. The patient completed an additional 2 week course of IV antibiotic therapy upon discharge. Mesenteric duplex ultrasonography performed 14 months post-operatively showed healthy celiac and SMA vasculature.

Case 5

A 61-year old female patient presented to an outside facility with severe abdominal pain. She became hypotensive and unresponsive in the emergency department resulting in intubation. The patient was transferred to our tertiary care facility and was stabilized with volume and blood product resuscitation in the intensive care unit. Her past medical history was insignificant for trauma, infections, or arterial disease that would predispose her to SMAA formation. CT imaging demonstrated a contained rupture from a 1.5 cm SMAA associated with a large retroperitoneal hematoma medial to the left kidney. On emergent laparotomy it was discovered she had a large hematoma of the transverse mesocolon. The inflow to the SMA was isolated at the base of the transverse mesocolon. We found a proximal branch off the SMA that was feeding the aneurysm. An Endo-stapler was used to fire a vascular, 1 mm staple height, cartridge to seal in the in-flow,

and then out going branches were dissected and ligated. The aneurysm was resected. Again, there was no need for revascularization or end organ resection. Post-operatively the patient was unable to overcome the insult from the initial hemorrhagic event. Repeat CTA did not show evidence for ongoing hemorrhage, and she underwent emergent bedside laparotomy in the surgical intensive care unit which did not reveal ischemic nor necrotic bowel. She remained coagulopathic, acidotic and died from multiorgan failure.

Discussion

Given the rare and often emergent presentation of SMAA, the primary etiology aneurysm formation has been a topic of debate. While a primary mycotic etiology was described as the most common presentation for SMAAs previously[7], more recent studies have suggested atherosclerosis to be the most common causative factor (Stone 2003). However, in up to 50-80% of cases, there is no identifiable cause, categorizing these SMAAs as cryptogenic in origin[3,8]. Similarly, 3 of our 5 cases did not have an identifiable etiology and were diagnosed as cryptogenic aneurysms. The pathogenesis and natural course of aneurysmal processes are well studied in aortic aneurysms[9], but have yet to be elucidated in the context of the SMA specifically. Hyperdynamic flow through the SMA in the presence of other visceral artery occlusions can cause aneurysm formation. Only the first case had an occluded celiac from compression by the SMAA. In the remaining cases no visceral artery occlusion was found to suggest this as an etiology. Aneurysmal degeneration of atherosclerotic disease may account for cryptogenic SMAAs. Samples were sent for histologic review in four of our cases – two cases (cases 3 and 5) were classified as ‘infectious psuedoaneurysms’, one cases (case 2) as ‘pseudoaneurysm with intimal thickening and adventitial scarring’, and the last (case 1) with no

definitive diagnosis, though significant atherosclerosis was noted. No case had histologic evidence of dissection, connective tissue disorder, nor vasculitis.

SMAAs occur most commonly in adults and have a male predominance[2], however in our short series, 4 of the 5 cases were female. While not represented in our series, case reports in relatively younger patients have pointed to more rare etiologies of aneurysm formation in the SMA including rheumatic endocarditis (with superimposed subacute bacterial endocarditis)[10], Brucella endocarditis[11], and suppurative adenitis in a patient with no other clear source of septic emboli[12]. Traumatic SMAA has also been reported[13]. Less common etiologies include Behcet's disease[14], Takayasu's arteritis[15], segmental arterial mediolysis[16,17], Ehlers-Danlos[18], and fibromuscular dysplasia[19]. Just as in adults, the pediatric population can also be affected by mycotic aneurysms, though rarely reported. While SMAA may present with preceding weight loss, decreased appetite, or as a sequelae of chronic inflammation such as pancreatitis, only one patient (case 3) in our series had such symptoms (reporting weight loss in the 2-3 months prior to aneurysmectomy). CTA is the primary modality for diagnosis, as it allows for simultaneous evaluation of the aneurysm as well as the other mesenteric vessels.

Mycotic aneurysms are most often caused by local degeneration of the arterial wall secondary to infection. Mycotic pathogenesis can be initiated by a) contiguous infection, b) hematogenous spread, c) septic emboli, typically in the context of infective endocarditis, or d) direct, contaminated arterial puncture, e.g. in the setting of non-sterile hospital conditions or IVDA[20,21]. Definitive treatment of the mycotic SMAA must address the aneurysm as well as treat with long term antibiotics if infection is found. A complete workup and effort to identify occult infection should be undertaken so as to not miss a distant source of seeding[20,22] It is for this reason it is our practice to approach SMAAs with open surgical therapy. While endovascular

therapies have been increasingly reported, we find this to be suboptimal management without definitively ruling out infection with direct cultures or operative examination. In our series, two of our patients had infective etiology and required long-term antibiotics. In case #4 the patient presented without leukocytosis, had negative blood cultures, and negative intraoperative cultures. However, with a history of genital infections and IVDA our suspicion was high. Upon operative exploration an inflamed and purulent aneurysm sac was discovered, resulting in treatment with long-term antibiotics.

According to the recently published Society of Vascular Surgery guidelines[6], repair is recommended for all SMAAs, whether a true aneurysm or a pseudoaneurysm. Indeed, this pathophysiology has not been well studied, and yet the outcomes of ruptured aneurysms are often severe. In this context and in line with these society recommendations, our department will recommend operation on a SMAA regardless of size, in the appropriate clinical context.

Several treatment strategies have been reported for SMAA. Open surgical approaches have been the mainstay of treatment of SMAAs. Endovascular techniques including stents and coil embolization have been reported for cases with[23-7] and without[14,28] rupture. Oechsle et al. used a percutaneous approach, injecting thrombin directly into the lumen, under ultrasound guidance, to achieve thrombotic occlusion of the aneurysm sac[29]. Zilun et al. published a series of 16 patients in which the majority of patients were electively treated with overlapping bare metal stents[23]. The majority of these patients did well, however two patients died; in one case due to stent thrombosis and in another due to delayed rupture of the SMAA. A recent systematic review of visceral artery aneurysms suggests mortality is actually similar between endovascular and open techniques[30]. There may be a role for endovascular treatments in patients with multiple medical co-morbidities and optimal lesions, and indeed, the recently published Society of

Vascular Surgery guidelines recommend an "endovascular-first approach to all SMAAs if anatomically feasible"[6]. These guidelines further suggest, however, that an SMAA that extends beyond the proximal few centimeters of the ostium will include important branches that must be maintained, and as such, open surgery is recommended more strongly. As this was the case in our experience, to avoid endovascular coverage of these branches, we opted for open repair. The majority of SMAAs are saccular aneurysms, have an increased risk of rupture, and are associated with mycotic etiology. Our practice is to treat each case definitively with resection and long-term antibiotics where infection is confirmed. Our approach has been via laparotomy to maximize our ability to gain vascular control, although there have been reports of laparoscopic resections of SMA aneurysms[31]. Direct surgical approaches also allow the opportunity to address end organ perfusion simultaneously.

Revascularization following aneurysmectomy is an important consideration in every case. The statuses of the celiac axis and inferior mesenteric artery are pre-operatively evaluated with CTA in each case. Intra-operatively, about 20-30 minutes following repair of the aneurysm, the bowel is re-assessed for its color and a palpable pulse and/or a Doppler signal at the mesenteric border to help determine whether revascularization is warranted. Other adjuncts, such as fluorescein injection and evaluation with a Wood's lamp can be considered as well.

Rupture is the feared complication of SMAA. An accurate estimate of the incidence of rupture is difficult, given the predominance of single case reports in the literature. In the largest study of 21 cases, 8 (38%) presented with rupture;[3] (Stone 2002) the operative mortality rate of these ruptured cases was 37.5%. In our institutional case series, we had a 100% mortality due to ruptured SMAA. There is a 15% reported mortality for surgical intervention of non-ruptured

SMAAs[32]. In our experience, patients we treated for SMAA who were not ruptured had a 0% mortality and no long-term morbidity due to our treatment.

Our approach to follow-up involves a mesenteric duplex study and initial clinic visit within 1 month, and repeated ultrasound evaluation and clinical assessment afterwards. Employing a high index of suspicion, patients are questioned about diet, weight loss, food-fear, nausea, abdominal pain, and any positive finding, especially if in the context of a borderline or positive mesenteric duplex, warrants a CTA and further work-up.

We described multiple saccular aneurysms involving the branches or main trunk of the SMA that were successfully treated with resection. When aneurysms involve branches of the main SMA, in our experience the need for reconstruction was limited, as there is significant collateralization through parallel circuits from the celiac, inferior mesenteric artery, and from the SMA itself. The limited role of resection in this scenario is encouraged by our case series as we did not need to resect bowel in any case that involved only aneurysmectomy. At follow up, all survivors have good bowel function and recovered fully from the surgical intervention.

Conclusion

SMAAs are a rare disease that require urgent management to avoid the often fatal consequences of rupture. Aneurysmectomy, with or without reconstruction, can treat the majority of SMAAs with good outcome, whether they involve the main trunk or the branches. As the experience in identifying SMAAs, diagnosing the etiology, and providing optimal treatment grows, devastating outcomes from aneurysm rupture may be better prevented.

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Figure 1: 3.2 cm SMA aneurysm from a branch of the Proximal SMA

Figure 2: Proximal SMA aneurysm at a branch point repaired with an interposition saphenous vein graft

Figure 3: CT imaging showing saccular aneurysm of the ileocolic branch of the SMA

Table 1: Clinical Characteristics

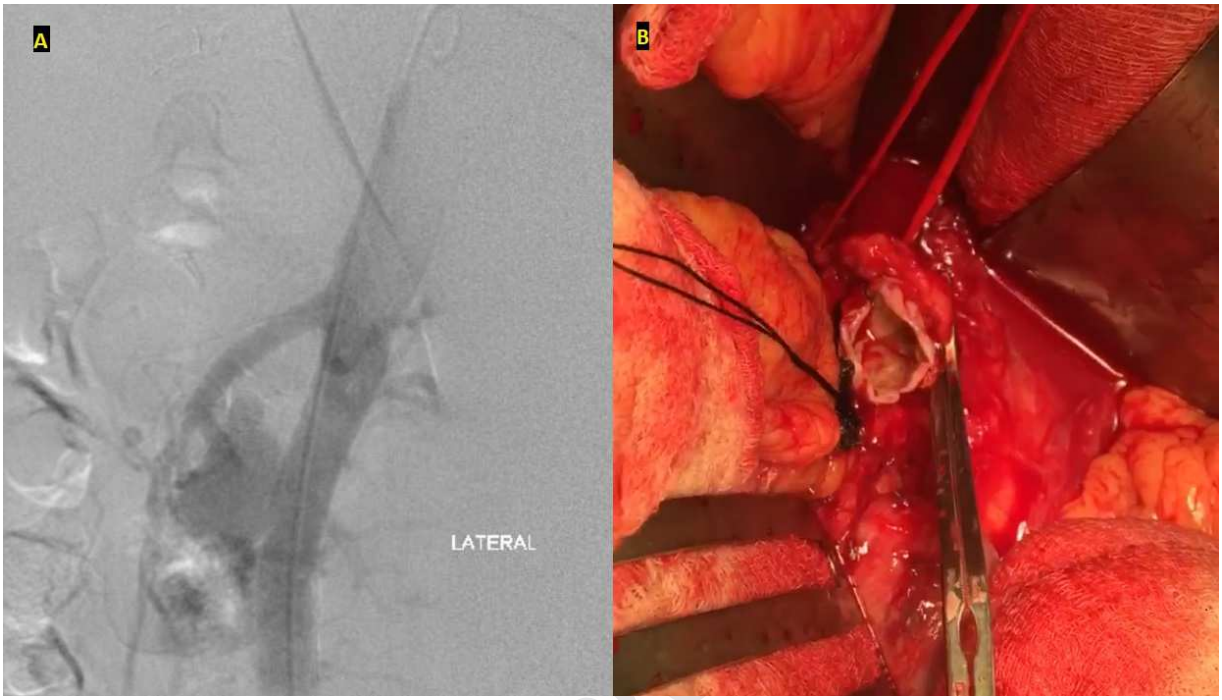
Case #	Age	Sex	Pertinent History	SMA Branch Point	Presentation	Size (cm)
1	75	F	None	Yes	Incidental Finding	3.5
2	54	F	None	Yes	Symptomatic (Back Pain)	1.5
3	74	F	Endocarditis	Yes	Symptomatic (RUQ pain)	1.2
4	70	M	IVDA	Yes	Incidental Finding	2
5	61	F	None	Yes	Rupture	1.5

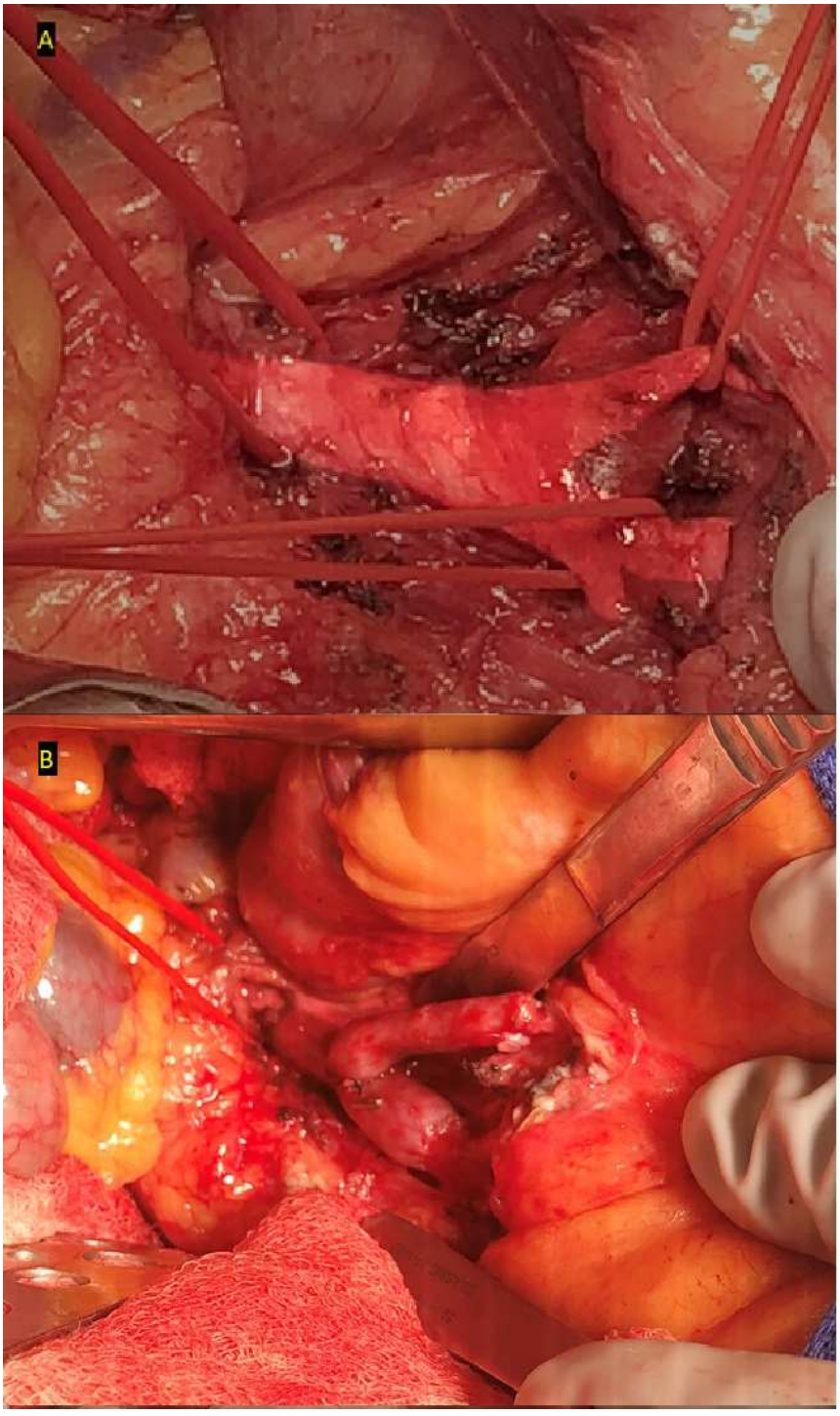
IVDA: Intravenous Drug Abuse; *RUQ:* Right Upper Quadrant; *SMA:* Superior Mesenteric Artery

Table 2: Interventions and Outcomes

Case #	Repair	Bowel Resection	Long Term Antibiotics	Outcome
1	PTFE Aorto-Common Hepatic Artery Bypass and Aneurysmectomy	No	No	Patient Bypass > 30 days
2	Aneurysmectomy and Bifurcated Reversed GSV Graft	No	No	Patient Bypass > 30 days
3	Aneurysmectomy	No	Yes	None
4	Aneurysmectomy	No	Yes	None
5	Aneurysmectomy	No	No	Mortality

GSV: Greater Saphenous Vein; *PTFE:* Polytetrafluoroethylene

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