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Vascular Interventions *Case Report*

Management of Visceral Mycotic Pseudoaneurysms

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ABSTRACT

Visceral arterial pseudoaneurysms are rare, however, if left untreated are associated with high rates of morbidity and mortality. Pseudoaneurysms typically develop as a result of trauma, infection, or underlying inflammatory disease. While management can be dictated by the size and etiology, clear guidelines are not well established, as there are no randomized trials to compare patient outcomes. Up to this point, management has involved antibiotic therapy, surgical debridement, and only more recently, endovascular approaches. We present a case of a 52-year-old male who postoperatively was found to have new liver, mesenteric, and splenic pseudoaneurysms which were all successfully treated through endovascular approach.

Keywords: Embolization, Hepatic pseudoaneurysm, Mesenteric pseudoaneurysm, Mycotic, Pseudoaneurysm, Splenic pseudoaneurysm, Visceral pseudoaneurysm

INTRODUCTION

Pseudoaneurysms typically develop as a result of trauma, infection, or underlying inflammatory disease.[1-5] Mycotic pseudoaneurysms are caused by infection of the arterial wall leading to wall destruction and an unstable arterial outpouching. As opposed to true arterial aneurysms, pseudoaneurysms do not contain all three layers of the arterial wall, therefore, making them more prone for rupture.[6] The previous approach to management of mycotic pseudoaneurysms has been antibiotic therapy and surgical debridement when possible.[7] Recently, endovascular approaches have emerged. However, evidence of success has been solely based on case reports rather than long-term data. There has been more data describing management of aortic aneurysms, showing that open repair is associated with greater morbidity than endovascular approaches.[8] However, ideal treatment for pseudoaneurysms of medium sized visceral vessels is less clear.

CASE REPORT

A 52-year-old male was admitted after routine follow-up imaging showed new hepatic, proximal splenic, and mesenteric pseudoaneurysms. Relevant clinical history included the patient undergoing a left nephrectomy 3 months earlier. Surgical pathology showed clear cell renal cell carcinoma and underlying necrotizing crescentic glomerulonephritis indicative of granulomatosis with polyangiitis (GPA). Postoperatively, he was started on rituximab. Approximately 2.5 months following surgery, the patient underwent an abdominal computed tomography (CT) due to pain which showed non-specific hypodensities in the spleen and liver most consistent with areas of

infarction. He was then started on apixaban. His outpatient 3-month follow-up imaging was then obtained, which showed new focal areas of enhancement [Figure 1] within the small bowel mesentery measuring 1.8 cm, in between the spleen and the stomach measuring 3.1 cm, and within the left hepatic lobe measuring 1.6 cm, most consistent with arterial pseudoaneurysms. Due to the prior evidence of infarctions, concern was raised for a central source of emboli. Echocardiogram and infectious work up were then ordered. The patient was then evaluated by surgery and found not to be a candidate for surgical intervention due to the multiple locations of pseudoaneurysms throughout the viscera. Interventional radiology (IR) was consulted for urgent management of new visceral pseudoaneurysms.

Due to recent intake of apixaban, radial access on hospital day 1 was elected for intended visceral pseudoaneurysm embolization. During this intervention, the left segment two hepatic arterial pseudoaneurysm was embolized using 40% of a vial of 300–500 μm Embospheres (Merit Medical Systems, Inc. South Jordan, UT, USA) and Concerto EV3 0.018" detachable coils (Medtronic USA, Inc. Minneapolis, MN, USA). Post-embolization angiography displayed sluggish flow within the vessel. Angiograms of the splenic and superior mesenteric arteries at the time did not show abnormally filling vessels. The patient was then brought back to IR on hospital day 2 and right common femoral access was obtained. Using fluoroscopy and CT, the jejunal branch supplying the mesenteric visceral pseudoaneurysm [Figure 2] was isolated using a straight Progreat 2.4Fr microcatheter (Terumo Medical Co. Shibuya City, Tokyo, Japan) and Fathom 0.014" microwire (Boston Scientific, Marlborough, MA, USA). The outflow vessel was selected and the pseudoaneurysm was coiled across the outflow and inflow vessels using 0.018" Concerto EV3 detachable coils. Coil embolization was performed because there was a single outflow vessel which could be selected allowing coiling across the PSA neck. Coils were also felt to be the most safe because at this level of the mesenteric arcade, coil embolization allowed small bowel perfusion through collaterals distally. Post-embolization angiogram showed good collateral perfusion to the bowel distal to the embolization coils.

Attention was then drawn to the pseudoaneurysm at the level of the splenic artery [Figure 3]. Due to high splenic arterial flow, angiography did not demonstrate pseudoaneurysm filling, however, there was persistent flow seen on intra-procedural delayed CT imaging. The distal splenic artery was selected using a 4Fr Kumpe catheter (Cook Medical, Bloomington, IN, USA) and Progreat 2.4Fr microcatheter and Fathom microwire. Distal splenic angiogram showed no arterial filling, therefore, directing our attention to the more proximal splenic artery branches. Proximal splenic arterial branches were selectively catheterized until the pseudoaneurysm filling artery was identified. Due to evidence of pseudoaneurysm collateral filling through short gastric arteries which could not be selected, the pseudoaneurysm was embolized using TRUFILL n-Butyl Cyanoacrylate: Ethiodized Oil 1:2 ratio mixture (CERENOVUS, Fremont, CA, USA). Liquid embolic was used in this location to ensure all short gastric contributing channels would be filled and because low risk associated with liquid embolic traveling more distally toward the end splenic artery.

Finally, the left hepatic and segment III hepatic arteries were selected using a new Progreat microcatheter and Fathom microwire [Figure 4]. Angiogram of the left hepatic artery revealed persistent pseudoaneurysm filling, which was then embolized using 300–500 μm Embospheres and fibered Nester coils (Cook Medical, Inc. Bloomington, Indiana, USA). Fibered Nester coils were used because non-fibered coils used 1 day prior by a different provider did not create an adequate seal. The patient tolerated the procedure well.

Following IR procedure, the patient was taken for trans-esophageal echocardiogram which showed multiple mobile vegetations on the atrial surface of the anterior mitral valve and severe mitral regurgitation. The patient was then transferred to an outside hospital for urgent cardiothoracic evaluation. Blood cultures revealed *Streptococcus* bacteremia at time of transfer.

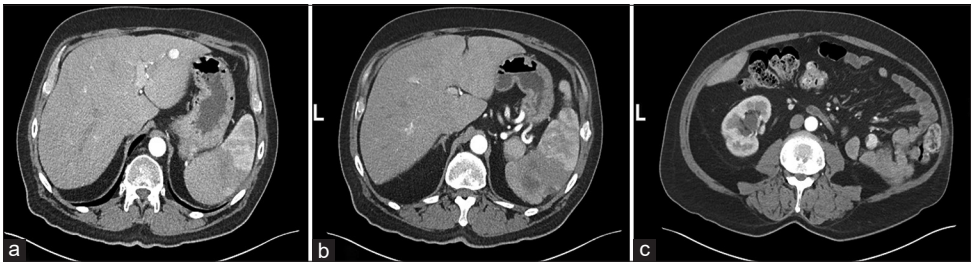

Figure 1: A 52-year-old male with a history of granulomatosis with polyangiitis and left nephrectomy 3 months earlier who presents for routine post-operative follow-up. Contrast-enhanced computed tomography abdomen shows new focal areas of enhancement in (a) the left hepatic lobe, (b) between the spleen and the stomach, and (c) in the small bowel mesentery most consistent with new pseudoaneurysms.

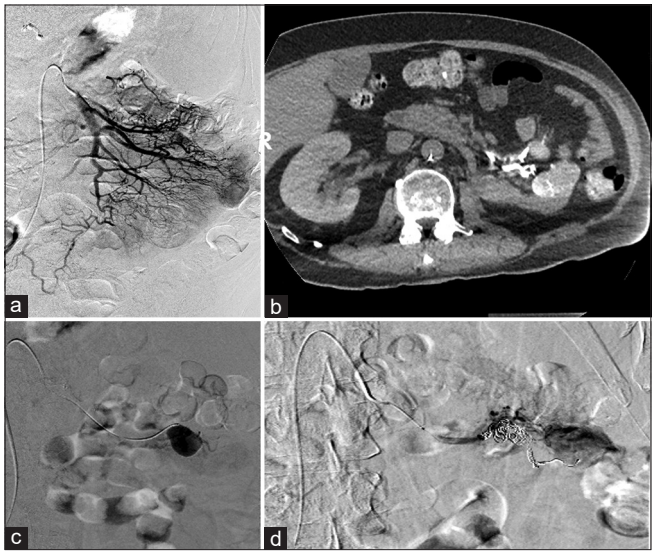

Figure 2: A 52-year-old male with a history of granulomatosis with polyangiitis and left nephrectomy 3 months earlier presenting for management of visceral pseudoaneurysms (PSAs). (a) Digital subtraction superior mesenteric angiogram shows no definite filling of the jejunal mesentery PSA. (b) CT during catheter injection into the third jejunal branch artery shows delayed PSA filling. (c) Superselective jejunal branch angiogram shows filling of the PSA as well as a small outflow vessel. (d) Outflow vessel was successfully selected and PSA embolized using coils.

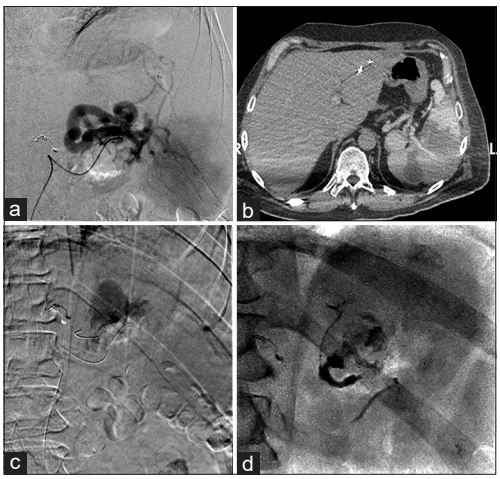

Figure 3: A 52-year-old male with a history of granulomatosis with polyangiitis and left nephrectomy 3 months earlier presenting for management of visceral pseudoaneurysms (PSAs). (a) Digital subtraction splenic angiogram did not show a clear origin of the pseudoaneurysm feeding vessel. (b) Intraprocedural computed tomography during contrast injection into the proximal splenic artery showed persistent flow within splenic PSA. (c) Digital subtraction angiogram after selecting a small inflow vessel arising off the proximal splenic artery showed a large PSA with outflow vessels through short gastric arteries. (d) PSA was successfully embolized using TRUFILL liquid embolic.

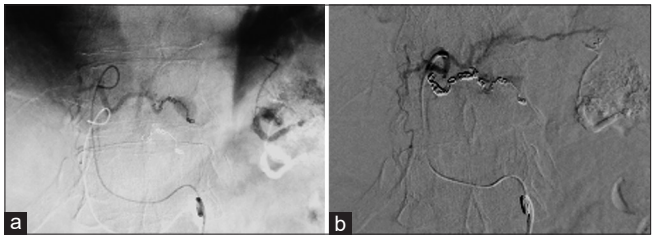

Figure 4: A 52-year-old male with a history of granulomatosis with polyangiitis and left nephrectomy 3 months earlier presenting for management of visceral pseudoaneurysms (PSAs). (a) Superselective hepatic segment III angiogram showing persistent flow within the PSA that was previously embolized with particles and coils. (b) Following embolization using particles and fibered coils, segment III postembolization angiogram showed no further filling of the PSA.

At the outside hospital, the patient underwent mitral valve replacement. Per record review, 1 week following the procedure, CT head was performed following mental status changes. Imaging showed acute intraparenchymal hematomas of the left parietal and occipital lobes. CT angiogram of the head was performed and showed multiple scattered small aneurysms of the distal arteries. Two aneurysms were present in the left parietal lobe, one in the right parietal lobe, and one in the right occipital lobe. The patient then underwent neurointervention where the right common femoral artery was accessed. The 6-Fr BENCHMARK guide catheter (Penumbra, Inc. Alameda, CA, USA) telescoping over the Berenstein angled diagnostic catheter (Merit Medical Systems, Inc. South Jordan, Utah, USA) and 0.035" Glidewire (Terumo Medical Co. Shibuya City, Tokyo, Japan) were advanced to selectively catheterize the left internal carotid artery and obtain three-dimensional angiography. A distal left middle cerebral artery (MCA) was identified correlating to the parieto-occipital hemorrhage seen on CT. Using a Synchro soft microwire (Stryker Coorporation, Kalamazoo, MI, USA) and Aristotle 18 microwire (Scientia Vascular, West Valley City, UT, USA) inside the 5 French Sofia intermediate catheter (Microvention, Aliso Viejo, California, USA); it was possible to advance the catheter past the ophthalmic segment. Under high zoom magnification and continuous fluoroscopy guidance, the Scepter mini balloon microcatheter (Microvention, Aliso Viejo, California, USA) telescoping over the hybrid microwire were advanced to the distal branch of the inferior division of the left MCA containing the mycotic aneurysm. Successful embolization was performed using the Onyx 34 (Medtronic, Inc. Minneapolis, MN, USA) and confirmed by final angiography.

The patient recovered and was discharged approximately 2 weeks later to follow-up with neurology, neurosurgery, speech pathology, cardiology, infectious disease, and rheumatology. The patient is now doing well at 6 months post-operative [Figure 5].

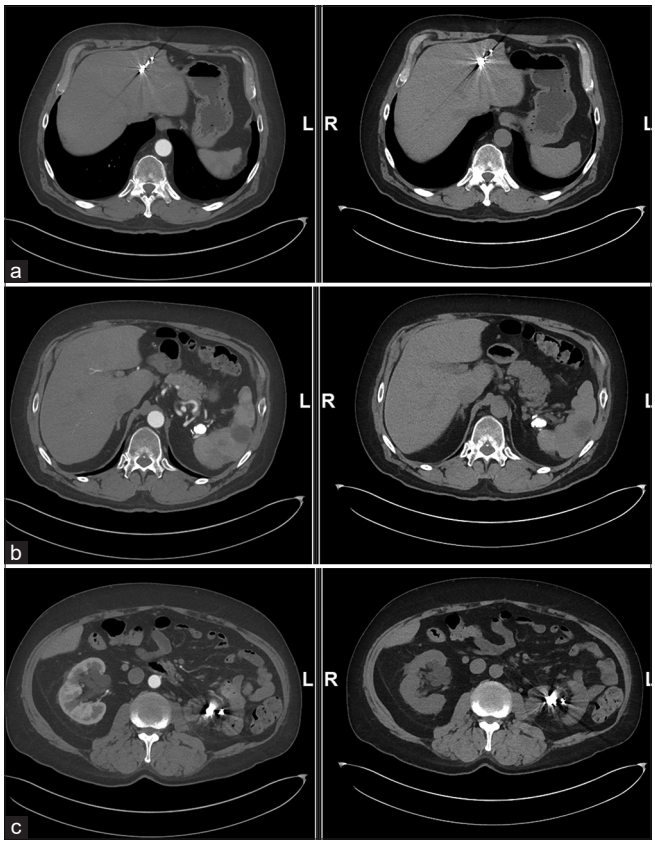

Figure 5: A 52-year-old male with of granulomatosis with history of polyangiitis, left nephrectomy and multiple visceral and brain mycotic pseudoaneurysms status post-endovascular embolization. Follow-up computed tomography (arterial phase on left, delayed phase on the right) 6 months after embolization shows (a) no pseudoaneurysm filling in the left hepatic lobe, (b) splenic artery, or (c) mesentery.

DISCUSSION

We describe a case of a patient who had undergone a left nephrectomy in the setting of underlying small vessel vasculitis (GPA) and whose symptoms of active infection were masked due to immunosuppression. Because visceral mycotic pseudoaneurysms are very rare and symptoms are non-specific, there must be high suspicion on patient evaluation and imaging. In symptomatic patients, clinical presentation may include pain at the site of the pseudoaneurysm, bleeding, ischemia (if source s septic emboli), fevers, etc. Imaging findings can include saccular/eccentric aneurysmal outpouching of the involved artery, adjacent soft-tissue inflammation, and surrounding foci of gas and/or fluid. Cross-sectional multiphase imaging is the most specific; however, peripheral pseudoaneurysms can also be evaluated with Doppler ultrasound imaging.

This patient's development of bacterial endocarditis likely stemmed from chronic immunosuppression in setting of rituximab usage to treat his underlying autoimmune inflammatory disorder. This systemic infection then led to septic emboli, depositing bacteria along arterial wall, which then led to the formation of multiple multifocal visceral mycotic aneurysms. Symptomatic peripheral mycotic aneurysms are very rare complication of infective endocarditis, one study reporting prevalence of 1.9% (18/922) of infective endocarditis cases.[9] Even more rare was associated intra-abdominal visceral aneurysms, occurring in 0.1% (1/922) of patients, and associated intracranial hemorrhage occurring in 0.65% (6/922) of patients.[9]

The previous approach to management of mycotic pseudoaneurysms has been antibiotic therapy and surgical debridement when possible.[7] Choice of antibiotics is usually broad-spectrum antibiotics initially which would then be further guided by the results of the blood cultures. There are no established guidelines for the duration of antibiotics; however, factors such as implanted devices, state of immunity, and location of infection should be considered. In some instances, inflammatory markers can be followed to assess for adequate response to medical management. In surgical candidates, resection of an infected nidus with vascular reconstruction has been described as the mainstay management to prevent further infection seeding. In non-surgical candidates, endovascular approaches have previously been described; however, evidence of treatment success has been solely based on case reports rather than long-term data. There have been more recent data describing management of aortic aneurysms, showing that open repair is associated with greater morbidity than endovascular approaches.[8] Ideal treatment for pseudoaneurysms of medium sized visceral vessels is less clear and can only be extrapolated from case series and aortic repair data. In our case, we have been successful at excluding the pseudoaneurysms through endovascular approach and now have a 6 month follow-up showing no recurrence.

CONCLUSION

Despite challenges associated with treating mycotic pseudoaneurysms, our embolization attempts were successful at excluding the mycotic pseudoaneurysm sacs without compromising distal flow to end organs. Since discharge, the patient has been free of gastrointestinal events and doing well on medical management, pointing to a useful role of endovascular interventions in management of visceral mycotic pseudoaneurysms.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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