



Hybrid Surgery to Treat Multiple Visceral Aneurysms Secondary to Polyarteritis Nodosa

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A 57-year-old woman presented to vascular surgery clinic with visceral artery aneurysms that were incidentally detected during regular check-up. Imaging studies revealed occlusion of the celiac axis and severe stenosis of the superior mesenteric artery and 3 aneurysms along the posterior and inferior pancreaticoduodenal arteries, as well as the right gastroepiploic artery. Endovascular embolization of all aneurysms was rejected because of the risk of hepatic ischemia. These complicated lesion caused by polyarteritis nodosa were successfully treated using a hybrid operation with coil embolization, aneurysm resection, and antegrade aorto-celiac-superior mesentery artery bypass.

Key Words: Aneurysm, Arterial occlusive diseases, Polyarteritis nodosa

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INTRODUCTION

A visceral artery aneurysm (VAA) is defined as an aneurysm originating from the celiac, superior and/or inferior mesenteric arteries or their branches. It is an extremely rare condition with a reported incidence of approximately 0.01%-2% [1]. The most common site of a VAA is the splenic artery, which accounts for approximately 60% of VAAs, followed by the hepatic artery (20%), the celiac trunk (5.5%), superior mesenteric artery (SMA) (4%), gastric and gastroepiploic arteries (4%), intestinal arteries (3%), and the pancreaticoduodenal arteries (PDAs) (2%). The development of VAAs can be attributed to various etiologies—the most common cause being atherosclerosis. VAAs can develop secondary to vasculitis that is noted in patients presenting with polyarteritis nodosa (PAN), Takayasu arteritis, and Kawasaki disease [2].

We describe a patient who presented with multiple VAAs and steno-occlusion of the celiac trunk and the SMA, which

were treated using hybrid surgery. The diagnosis of PAN was confirmed histopathologically.

CASE

A 57-year-old woman presented to vascular surgery clinic with VAAs that were incidentally detected during regular check-up. She did not relate any specific medical history other than hypertension. Computed tomography (CT) revealed occlusion of the celiac axis and severe stenosis of the SMA and 3 aneurysms along the posterior and inferior PDAs, as well as the right gastroepiploic artery (RGEA) (Fig. 1).

She denied all symptoms suggestive of intestinal angina or vasculitis. Laboratory work-up revealed white blood cells 5,440/mm³ (reference range, 4,000-10,000/mm³), erythrocyte sedimentation rate 30 mm/h (reference range, 10-20 mm/h), blood urea nitrogen 9 mg/dL (reference range, 10-26 mg/dL), creatinine 0.61 mg/dL (reference range, 0.7-

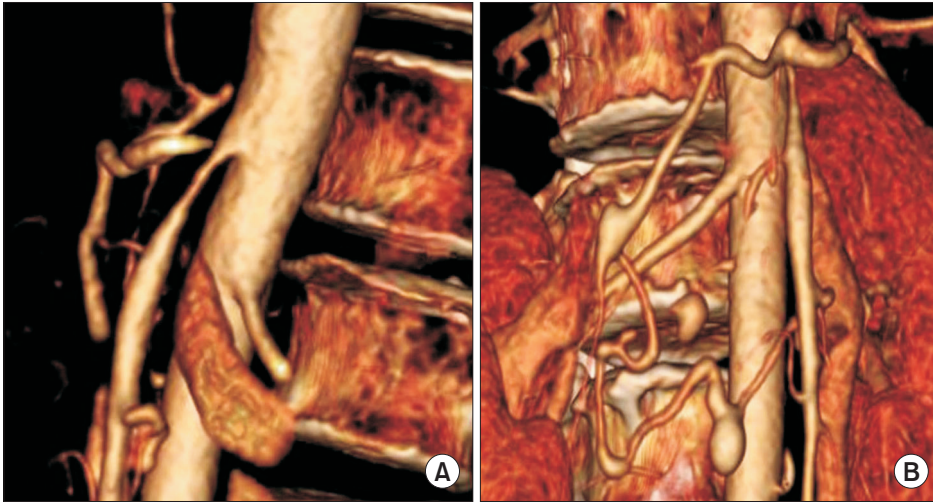


Fig. 1. (A) Near-total occlusion of the celiac trunk and severe stenosis of the superior mesenteric artery. (B) Multiple aneurysms of the right gastroepiploic artery and posterior and inferior pancreaticoduodenal arteries.

1.4 mg/dL), C-reactive protein 0.09 mg/L (reference range, 0–0.5 mg/L), and her anti-neutrophil cytoplasmic antibody (ANCA) was negative.

Considering the possibility of vasculitis, such as PAN, rheumatology consultation was performed to decide whether administration of systemic immunosuppressants would be required prior to surgery. The patient did not complain of any constitutional symptoms, nor any symptoms associated with the lesion. The lesion was well localized and considered to be chronic in nature. Therefore, systemic immunosuppressant administration was deferred. Endovascular embolization was rejected as a possible treatment option for the management of the 3 aneurysms because of the risk of hepatic ischemia secondary to the obliteration of the SMA-to-celiac collaterals. The posterior PDA aneurysm observed in a retropancreatic location was first treated using embolization with multiple coils and a glue-lipiodol mixture. Two days later, we performed open surgery via a midline incision, and she underwent an antegrade aorto-common hepatic artery-SMA bypass utilizing a 14/7 mm Dacron Y graft (Hemashield; Maquet, Rastatt, Germany), as well as an aneurysmorrhaphy of the RGEA aneurysm and aneurysmectomy of the inferior PDA aneurysm. Histopathology showed acute and chronic inflammatory cell infiltrates in the media and adventitia. Lymphocytic and neutrophilic infiltration as well as hyaline degeneration and fibrinoid necrosis were confirmed, and the patient was diagnosed with PAN. Postoperative CT showed a patent graft and disappearance of the visceral aneurysms (Fig. 2, 3). After confirming the diagnosis of PAN, the patient was treated with methotrexate (Yuhan Co., Seoul, Korea) at a dose of 10 mg daily for 6 days per week and prednisolone (Yuhan Co.) at a dose of 20 mg daily, which was eventually tapered to 2.5 mg daily.



Fig. 2. Postoperative computed tomography angiography showing the antegrade aorto-celiac-superior mesenteric artery bypass.

DISCUSSION

A VAA is a rare disease entity with an incidence of approximately 2%. Patients diagnosed with PAN, presenting with VAAs often show multiple sites of involvement [3]. In this patient, the PDA and the RGEA, which very rarely show the development of VAAs, were observed to show simultaneous involvement.

Usually, a VAA requires treatment only if it measures >2 cm, is symptomatic, or shows growth in size [4]. However, PDA aneurysms are associated with a high risk of rupture and therefore need immediate treatment following diagnosis regardless of their size. PDA aneurysms are known to be associated with celiac axis stenosis—it is hypothesized that

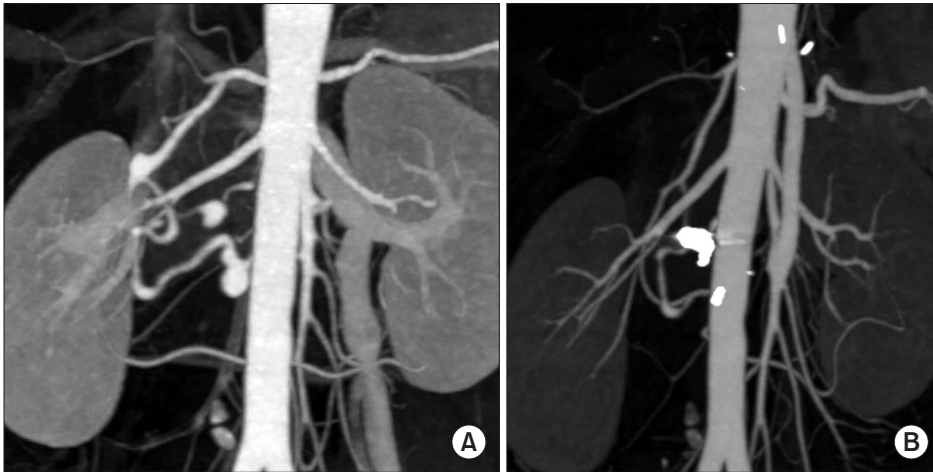


Fig. 3. Comparison of preoperative (A) and postoperative (B) computed tomography angiography.

an increase in collateral flow plays a role in the formation of these aneurysms [5].

Treatment of coexisting celiac axis steno-occlusion remains unclear in literature. However, in this patient, multiple PDA aneurysms were observed, and there existed a high risk of hepatic or duodenal ischemia. Therefore, antegrade aorto-celiac-SMA bypass was performed simultaneously.

Brocker et al. [5] have reported their practice recommendations for the treatment of co-existing celiac axis stenosis. Briefly, treatment of celiac axis stenosis should be initiated promptly if: 1) angiography demonstrates that the patient's anatomy is concerning for potential hepatic or duodenal ischemia, 2) the patient develops ischemic symptoms after initial definitive therapy, or 3) if the patient continues to demonstrate persistent symptoms.

GEA aneurysms are rare among splanchnic artery aneurysms and are associated with a high risk of rupture necessitating prompt treatment. Few cases have been reported in this regard; thus, definitive guidelines regarding indications for surgery/intervention have not yet been established. However, to date, surgery remains the treatment of choice for the management of these lesions [6].

Based on the Chapel Hill Consensus Conference 2012,

PAN is defined as necrotizing arteritis of medium or small arteries without the presence of glomerulonephritis or vasculitis in arterioles, capillaries, or venules, and a condition that is not associated with ANCA [7]. The disease spectrum ranges between single organ involvement and polyvisceral failure. Administration of glucocorticoids and cyclophosphamide forms the primary component of the treatment strategy to manage PAN. Milder forms are treated with only corticosteroids; however, cyclophosphamide is used concomitantly in patients showing critical organ involvement. The prognosis of PAN has improved in recent times owing to rapid and prompt diagnosis and more effective use of drugs. The 5-year survival rate for treated patients is approximately 80% [8].

In conclusion, multiple VAAs and celiac artery occlusion caused by PAN were successfully treated in our patient using a hybrid operation with coil embolization, aneurysm resection, and antegrade aorto-celiac-SMA bypass. Peripancreatic aneurysms should be treated promptly following diagnosis, and the treatment method should be specifically tailored in each patient based on the anatomy and the patient's need for revascularization.

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