中文題目：總肝動脈及脾動脈動脈瘤在一位嚴重型 A 型血友病病人上

英文題目：Common hepatic artery and splenic artery aneurysms in a patient with Severe Hemophilia A

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Introduction:

Hemophilia A is an inherited bleeding disorder that increases bleeding tendency and bleeding severity. Visceral artery aneurysm, a rare vascular pathology, has risks of rupture and, sudden death. Here we presented a case of visceral artery aneurysm in a patient with severe Hemophilia A.

Case Presentation:

A 48-year-old male with a history of Severe Hemophilia A and Type 2 diabetes mellitus presented to our emergency department because of recurrent upper abdominal pain, vomiting with coffee ground vomitus, and black stool passage. Panendoscopy was arranged, which revealed reflux esophagitis Los Angeles Grade A, hemorrhagic gastritis, and shallow gastric ulcer. After admission, he received intravenous Antihemophilic Factor VIII, Fc Fusion Prot (50IU/Kg) every 12 hours, proton pump inhibitor with esomeprazole, and tranexamic acid. Factor VIII inhibitor was undetectable. Abdominal sonography was arranged due to abdominal pain, which revealed two mass lesions with cystic content 7.1cm, 10.5cm over pancreatic body to tail. Abdominal computed tomography was arranged for further study, which revealed a large aneurysm about 15.3*7.5cm arising from celiac trunk with active contrast medium extravasation (Figure 1). Celiac angiography of 3 vessels (splenic, left gastric, and common hepatic artery) revealed two saccular vascular lesions, the smaller one, 1.3cm, at proximal common hepatic artery, and the larger saccular one, 7.1cm with daughter sac, at proximal splenic artery (Figure 2, left). Endovascular treatment with vascular plug embolization was performed (Figure 2, right). After procedure, we kept intravenous Antihemophilic Factor VIII, Fc Fusion Prot (50IU/Kg) every 12 hours and tranexamic acid. Followed Factor VIII activity was around 152~216%. Abdominal computed tomography after 14th days of vascular plug embolization revealed no active extravasation (Figure 3). The patient was discharged with intravenous Antihemophilic Factor VIII, Fc Fusion Prot (40IU/Kg) every 12 hours.

Discussion:

Visceral artery aneurysms (VAAs) are rare vascular pathologies with reported incidence rates of 0.01% to 0.2%[1].Aside from renal artery aneurysm, which have a separate cause, hepatic and splenic artery aneurysms are the most common forms of visceral artery aneurysms. Splenic artery and hepatic artery aneurysms treated with an endovascular approach and splenic artery and hepatic artery aneurysms treated with open surgery had no difference in short-term and long-term mortality, and overall mortality rates were low (<0.1%)[1].

Hemophilia A and B, the bleeding disorders inherited in a sex-linked fashion, are classified clinically into mild, moderate, or severe in severity. In severe type of hemophilia, spontaneous hemorrhage from early infancy can occur. The main clinical features of Hemophilia, especially in

severe type, includes hemarthroses and soft tissue hematomas. There are few case reports discussing about aneurysms in patients with hemophilia, and the incidence of aneurysms in patients with hemophilia is unknown. Here we presented a case of visceral artery aneurysm treated with vascular plug embolization in a patient with severe hemophilia A.

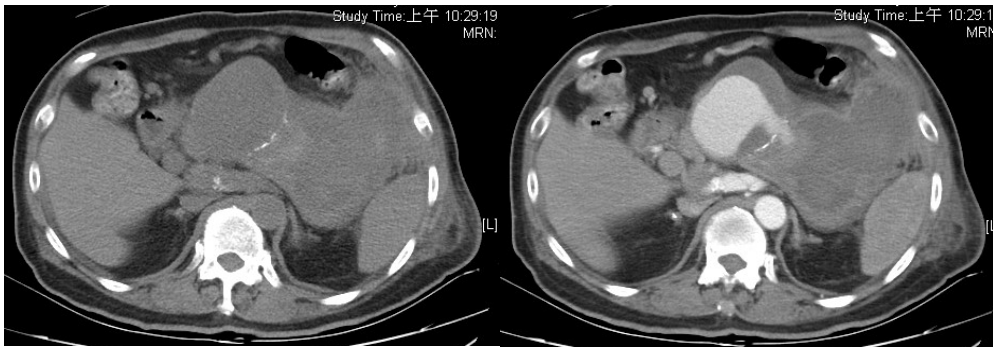

Figure 1: A large aneurysm about 15.3*7.5cm arising from celiac trunk with active contrast medium extravasation

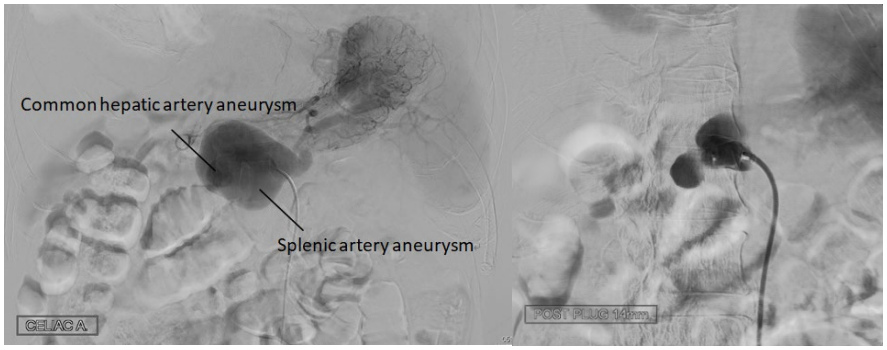

Figure 2: Aneurysms at proximal splenic artery and common hepatic artery (left), endovascular treatment with vascular plug embolization of splenic artery aneurysm (right)

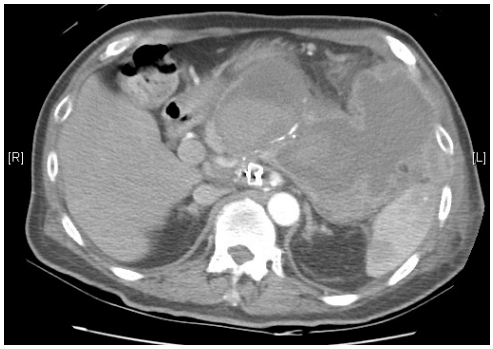

Figure 3: 14 days after vascular plug embolization, without active extravasation

Reference:

1. P. Barrionuevo et al. A systematic review and meta-analysis of the management of visceral artery aneurysms. J Vasc Surg. 2019;-:1-6