Pancytopenia and Massive Gastroesophageal Bleeding Due to Hypersplenism Treated with Partial Splenic Embolization

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Introduction: Portal hypertension may cause gastrointestinal complications; one of the most serious is a ruptured esophageal varices. Portal hypertension is also the main cause of hypersplenism, which in turn could lead to pancytopenia. Despite adequate therapy, some cases of hypersplenism could not be resolved. Partial splenic embolization (PSE) is an effective alternative method to treat this condition.

Method: We reported two cases of hypersplenism treated with PSE. The first case was a 10-year-old girl with pancytopenia and a history of recurrent esophageal ligation. The second case was a 32-year-old man with recurrent episodes of hematemesis for two years before admission.

Results: After the PSE procedure, the first patient's white blood cell and platelet doubled in one month after procedure and stable at follow-up three months later, with no complaint of hematemesis. The second patient's platelet doubled five days after the procedure. The first patient developed a complication of a splenic abscess, but after antibiotic administration and pus drainage, the condition was resolved.

Conclusion: PSE is an effective method to treat hypersplenism secondary to the hypertensive portal. Treatment goals successfully achieved include improvement in blood count and control of bleeding. There are risks following PSE, but with adequate treatment, it can be overcome.

Keywords: hypersplenism, partial splenic embolization, portal hypertension, pancytopenia
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INTRODUCTION

Portal hypertension may cause gastrointestinal complications such as esophageal varices, gastric varices, and hemorrhoids.¹ Acute variceal bleeding is perhaps the most severe consequence of uncontrolled portal hypertension and an increase in mortality.² In chronic liver disease, portal hypertension is a main cause of hypersplenism. Hypersplenism refers to a group of syndromes that involve peripheral cytopenia and splenomegaly of various causes.³⁻⁴

Despite adequate therapy, some cases of hypersplenism could not be resolved. Partial splenic embolization (PSE) is an effective alternative method to treat thrombocytopenia secondary to hypersplenism. PSE is normally reserved because of its potential complications, which are a post-embolization syndrome, splenic abscess, splenic rupture, and gastrointestinal bleeding.⁵ These cases are an example of the use of PSE as an effective

method to treat pancytopenia and massive gastrointestinal bleeding and its potential complication.

CASE ILLUSTRATION

First case.

A 10-year-old girl with a history of spleen enlargement since 3 years old, had recurrent episodes of hematemesis, have undergone endoscopy 17 times, and esophageal ligation three times in the last four years. She was hemodynamically stable (HR 92 bpm blood pressure 98/50). On examination, she was pale. Splenomegaly was found in schuffner IV. There was no enlargement of the lymph nodule. In the laboratory, the patient's hemoglobin, white cell, and platelet levels were 9.3 g/dL, $2.5 \times 10^9/L$, and $32 \times 10^9/L$, respectively. The international normalized

Figure 1. Result of esophagogastrosocopy 5 months before procedure. Thin arrow indicates esophageal varices grade 2. The white arrow head indicates mild hyperemic, gastropathic, erosive corpus mucosa.

ratio was 1.28, and the gamma-glutamyl transferase was 7 μ /L. The liver profiles and urinalysis were within normal limits. On the last endoscopy 5 months ago showed esophageal varices grade II caused by hypoplasia, suspected Banti's Syndrome (Figure 1). It was concluded that the pancytopenia was caused by hypersplenism secondary to portal hypertension.

Despite adequate treatment, there was limited improvement of the patient. She was referred for partial splenic embolization. Six days later, the procedure was performed. She was given the information about the procedure and the risk involved with it, that is, pain, bleeding, postembolisation syndrome, venous thrombosis, and splenic abscess. Written consent was taken. The hemoglobin, white cell, and platelet levels pre-procedure were 8.6 g/dL, 2.4×10^9 /L, and 30×10^9 /L, respectively. Three doses of 280 ml thrombocyte concentrate were given three hours before, during, and after procedure. Cefoperazone sulbactam 1.5 g was also given before the procedure.

Under general anesthesia, PSE was performed. The catheterization of the splenic artery

was achieved through the right femoral artery approach. Lower pole and mid pole branches of the splenic artery were selectively cannulated and injected with polyvinyl alcohol 500 μ m via a 5Fr Yashiro catheter. Postembolisation angiogram revealed an approximate total of 50% of spleen was embolized. No immediate complication was encountered during the procedure, and the patient's vital signs remained stable throughout.

Two days after the procedure, the patient developed a fever. Patient hemoglobin, white cell, and platelet levels were 9.1 g/dL, 13×10^9 /L, and 33×10^9 /L, respectively. Increased urine leukocytes were found. Thus fosfomycin 2 g was given. From urine culture was found enterococcus faecalis was found. Patient also developed complain of abdominal pain. Seventeen days after the procedure, abdominal USG showed a decrease in ecogenicity, suggesting splenic liquefaction. Abdominal CT also showed abscess caudal to the spleen. Therefore, the patient was diagnosed with splenic abscess. Laparotomy was performed on the patient for pus drainage and adhesiolysis. Ten days after laparotomy, abdominal USG showed smaller spleen, and liquefaction seems

Figure 2. Result of esophagogastrosocopy 4 months after procedure. Thin arrow indicates grade-1 esophageal varices. The white arrow head indicates mild esophagitis, mild gastritis, and repaired gastropathy.

Figure 3. Digital subtraction angiogram of the splenic artery. (a) Image shows splenic arteriogram pre-procedure. The thin arrow indicates the medial pole branch of the splenic artery. The white arrow head indicates lower pole branch of the splenic artery. (b) Image shows splenic arteriogram post procedure. Note that the medial and lower pole branch were embolized.

much reduced, with no sign leads to a splenic abscess. The culture showed growth of *Staphylococcus saprophyticus*. The last blood result showed hemoglobin, white cell, and platelet levels of 9.3 g/dL, $7.1 \times 10^9/L$, and $108 \times 10^9/L$, respectively. The patient then sent home.

Follow up was done four months after PSE. The patient no longer has complaints. Laboratory results showed hemoglobin, white cell, and platelet levels of 10.6 g/dL, $3.8 \times 10^9/L$, and $70 \times 10^9/L$, respectively. The liver profiles, albumin, and bilirubin levels were within normal limits. Endoscopy (Figure 2) showed esophageal varices grade I with resolved gastropathy.

Second case.

A 32-years-old man came to the emergency department with a chief complaint of hematemesis melena for one week before admission. This complaint occurred 2 years ago. The patient underwent ligation for esophageal varices six months ago. She was hemodynamically stable (HR 81 bpm BP 105/48). Physical examination showed hepatomegaly and splenomegaly. From the laboratory, the patient's hemoglobin, white cell, and platelet levels were 2.2 g/dL, $9.1 \times 10^9/L$, and $171 \times 10^9/L$, respectively. The international normalized ratio was raised, measuring 1.14. The renal, liver profile, electrolyte, and random blood glucose were within normal limits. HbsAg, Anti-HCV, Anti-HIV, and VDRL were all negative. Last abdominal MR seven months ago showed hepatomegaly on left lobe suggested chronic parenchymal liver disease, splenomegaly with portal hypertension, with no sign of stenosis or portal vein thrombosis. From pathology specimen showed partial degeneration of hepatocytes, portal fibrotic, suggest portal sclerosis, or obstructive portal venopathy. The patient was given 2500 ml of packed red blood cell and 500 ml of fresh frozen plasma. He was then referred for partial splenic embolization. Ten units of platelet and cefoperazone sulbactam 1 g

was given before the procedure. The hemoglobin, white cell, and platelet levels pre-procedure were 10.2 g/dL, $7.36 \times 10^9/L$, and $56 \times 10^9/L$, respectively.

Under general anesthesia, PSE was performed. The catheterization of the splenic artery was achieved through the right femoral artery approach. Lower pole and mid pole branches of the splenic artery were selectively cannulated and injected with polyvinyl alcohol 500 μ m via a 5Fr Yashiro catheter. Postembolisation angiogram revealed an approximate total of 50% of spleen was embolized (Figure 3). No immediate complication was encountered during the procedure, and the patient's vital signs remained stable throughout.

Five days after the procedure, the patient had no complaint. The vital sign was within normal limits. The hemoglobin, white cell, and platelet levels were 11.2 g/dL, $12.9 \times 10^9/L$, and $105 \times 10^9/L$, respectively. The patient then sent home. Two weeks after the procedure, the patient came to the clinic with no complaint. The hemoglobin, white cell, and platelet levels were 11.2 g/dL, $11.3 \times 10^9/L$, and $401 \times 10^9/L$, respectively.

DISCUSSION

T Pancytopenia refers to a disorder in which all hemoglobin, white blood cell, and thrombocyte are lower in counts than normal. One of the main causes of pancytopenia is secondary to hypersplenism.⁶ Hypersplenism is mainly caused by portal hypertension in chronic liver disease.³ Increased perfusion to spleen induces the overactive function of the spleen. Portal hypertension also causes gastrointestinal complications; one of the most serious is esophageal varices rupture. This condition could lead to death if not appropriately managed.²

In general, the two main treatments for varices are reducing portal pressure and endoscopic variceal ligation.² However, in these cases, despite adequate treatment, the patients developed recurrent variceal bleeding and no improvement in

blood function. PSE has been proposed as an alternative treatment modality for the management of hypersplenism with goals to improve blood function and reduce variceal bleeding. PSE is more favorable than splenectomy as it is generally a gentler approach and is associated with lower mortality rate.⁵

Splenic embolization for hypersplenism was first performed in the 1970s. After that many studies have been conducted to assess the effect on pancytopenia and variceal bleeding. In some studies, analysis reveals that 50 patients with an average of 2.4 bleeding episodes per year occurred before PSE, followed by an average of 0.48 bleeding episodes per year after PSE. The efficacy of splenic embolization for improving hematologic parameters has been shown in leukocyte count, red blood cell count, and platelet count after PSE. The suggested mechanism is PSE, which may reduce the splenic blood pool and sequestration. Another efficacies are the improvement in liver function, reduce hepatic encephalopathy, and increase the survival rate.⁷ Another study also showed that up to 80% splenic embolization is safe.⁸ In these cases, after 50% PSE, the first patient's white blood cell and platelet doubled in one month after procedure, and stable at follow-up three months later, with no complaint of hematemesis. The second patient's platelet doubled five days after the procedure compares to pre-procedure.

There are several risks associated with the procedure; one of the major is a splenic abscess. The splenic abscess has been documented occur in 6.8% of patient undergoing PSE. The previous study used second-generation cephalosporin or combination of ampicillin and gentamicin for intra-arterial antibiotics during PSE and showed a lower rate of splenic abscess formation.⁹ Treatment for splenic abscess include fine-needle aspiration, catheter drainage, to splenectomy. In the first case, the patient developed splenic abscess despite cephalosporin administration during the procedure, caused by *Staphylococcus saprophyticus*. Fosfomycin was given, and after laparotomy for drainage, the patient's condition got better.

Contraindication of PSE include the terminal stage of underlying disease and presence of ongoing infection, as it is associated with the formation of splenic abscess after procedure.⁷

PSE could be an alternative treatment modality for pancytopenia and esophageal varices bleeding secondary to portal hypertension and hypersplenism.¹⁰ This is the first report that showed PSE performed in Indonesia. This proves the efficacy of PSE for improving hematologic parameters and reduce variceal bleeding. There are risks following PSE, but with adequate treatment, it can be overcome.

CONCLUSION

The PSE could be an alternative treatment modality for pancytopenia and esophageal varices bleeding secondary to portal hypertension and hypersplenism.¹⁰ This is the first report that showed PSE performed in Indonesia. This proves the efficacy of PSE for improving hematologic parameters and reduce variceal bleeding. There are risks following PSE, but with adequate treatment, it can be overcome.

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