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Multiple splenic artery aneurysms in EHPVO patients: A case series

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Abstract
The most common visceral artery aneurysm is splenic artery aneurysm. Splenic artery aneurysm (SAA) occurs predominantly in multiparous women. The main risk factors are female gender, multiple pregnancies, atherosclerosis, hypertension, cirrhosis, and portal hypertension. SAAs are usually asymptomatic. Giant SAAs are more likely to cause symptoms and result in life-threatening complications. Multiple splenic artery aneurysms are very rare and only a few cases are reported. Available methods for the treatment of SAAs include endovascular techniques, open and laparoscopic surgical approaches. We present three cases of multiple splenic artery aneurysms diagnosed incidentally in patients with extrahepatic portal vein obstruction. All patients were treated simultaneously with aneurysm excision, distal pancreatosplenectomy and esophagogastric devascularization procedure. There was minimal morbidity and no mortality in our series.

Keywords: Splenic artery aneurysm, extrahepatic portal vein obstruction, distal pancreatosplenectomy, esophagogastric devascularization

Introduction
The most common abdominal artery aneurysm is the aorta followed by iliac arteries and splenic artery. SAA accounts for 50-75% of all visceral artery aneurysm cases [1]. True splenic artery aneurysms are rare and have a prevalence of 0.02-0.1% [2]. Splenic artery aneurysm is more common in females, especially during pregnancy with an increased risk of spontaneous rupture. Splenic artery aneurysms are usually single and < 2 cm in size. Giant aneurysms (> 2.5 cm) are quite rare with few cases reported till date [3]. Extrahepatic portal vein obstruction refers to the obstruction and cavernous transformation of the main portal vein with or without the involvement of the intrahepatic portal vein, splenic vein, or superior mesenteric vein. The incidence of SAA in patients with cirrhosis and portal hypertension is 7-20%. The incidence of portal hypertension in SAA is 20% [4]. However, multiple SAA in the clinical setting of portal hypertension is very rare. Despite their rarity, they are clinically of the utmost importance because of the occurrence of high-risk devastating complications like spontaneous rupture and bleeding.

Case Details
Case-1
A 36-years-old female patient presented with abdominal pain in the left hypochondrium, early satiety, no hematemesis and melena. Three years ago, patient had hematemesis and was diagnosed with Extrahepatic portal vein obstruction. The patient underwent endoscopic variceal band ligation for Grade III bleeding esophageal varices and was on secondary prophylaxis. An examination revealed moderate splenomegaly and no hepatomegaly. The complete blood count showed hemoglobin of 9.8gms/dl, a total count of 2800 /mm^3, and a platelet count of 62000/mm^3 with a normal liver function test. Peripheral smear report showed leukopenia and thrombocytopenia. Bone marrow reported reactive marrow with no abnormal cells. The upper gastrointestinal endoscopy revealed Grade I esophageal varices. Ultrasound of the abdomen and pelvis showed Portal cavernoma with multiple perisplenic, peripancreatic and lienorenal collaterals. The spleen measured 18cm. Contrast-enhanced computed tomography of the Abdomen and pelvis demonstrated a Portal vein replaced by multiple collaterals. Multiple dilated fusiform ecstatic segments (3 in numbers) were seen

along the entire course of the splenic artery, measuring 5.7×4.8 cm [Fig-1]. The patient underwent laparotomy. Intraoperatively there was a grossly enlarged spleen with multiple tortuous collaterals in the perisplenic, perigastric, and periportal regions. Splenic artery aneurysms of varying sizes, the largest one measuring 5×4 cm were present in the middle and distal parts. Splenic artery was ligated proximally. The distal portion of the pancreas was transected at the appropriate level. Distal pancreatosplenectomy along with aneurysm excision and esophagogastric devascularization was done [Fig-2 and Fig-3]. The postoperative period was uneventful. After 6 months of surgery, the patient had normal blood counts and the upper gastrointestinal scope showed no varices. The patient was symptom-free in subsequent follow-up.

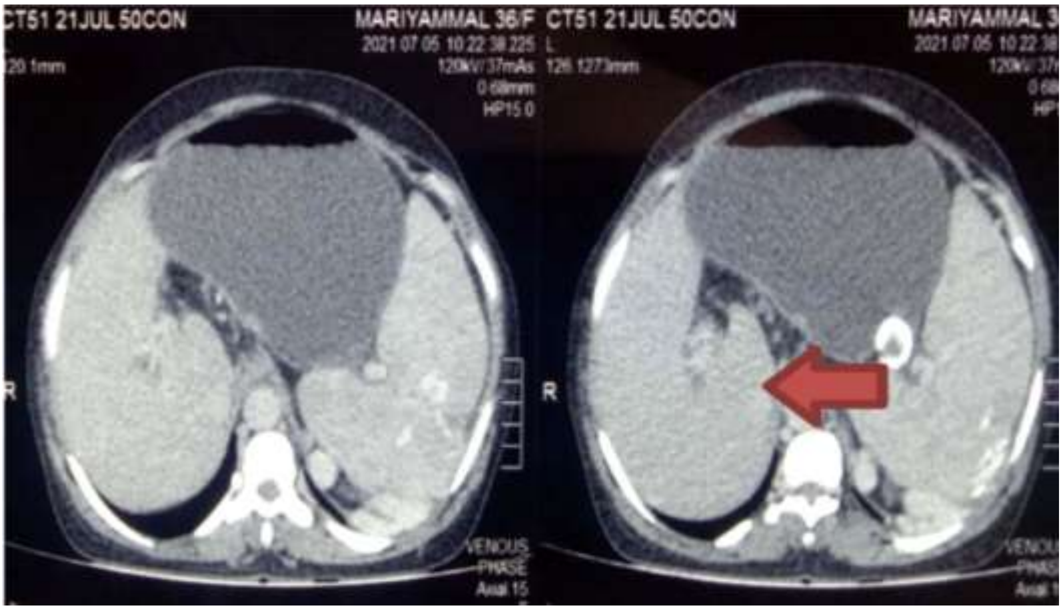

Fig 1: Contrast enhanced CT Abdomen & Pelvis: Axial cuts showing Splenic artery aneurysm with peripancreatic , perisplenic collaterals. (Arrow)

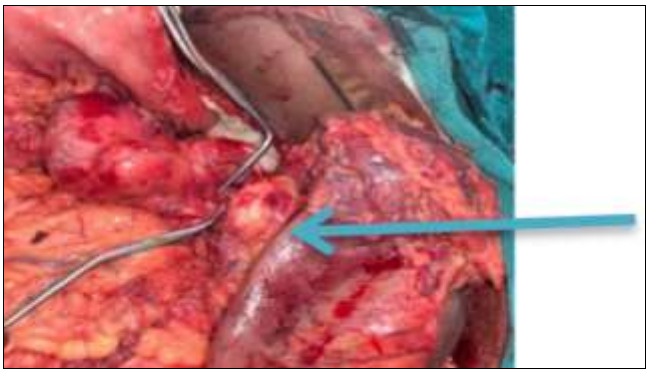

Fig 2: Multiple splenic artery aneurysm present along the splenic artery

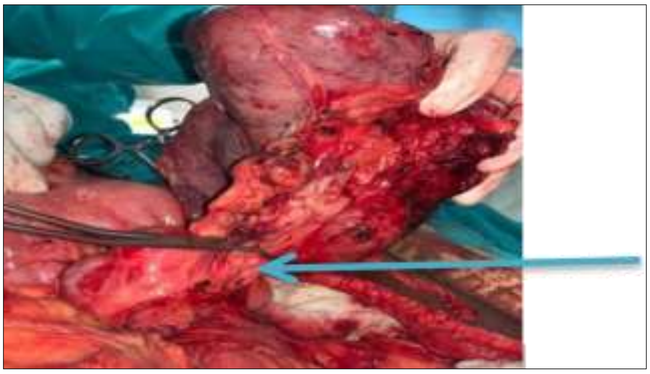

Fig 3: Distal pancreatosplenectomy with aneurysm excision

Case 2

A 46-years-old female patient presented with complaints of hematemesis. On examination, the patient was pale. Splenomegaly was present. Blood investigation showed hemoglobin of 5.6 gms/dl. After 3 units of packed cell transfusion, hemoglobin was increased to 9.8 gms/dl. The total count was 2700 cells/mm^3. The platelet count was 78000 cells/mm^3. Peripheral smear showed pancytopenia with no abnormal cells seen. Bone marrow reported as scattered hematopoietic elements.

Upper gastrointestinal scope findings revealed the presence of large fundal varices with no esophageal varices. Ultrasound of the abdomen and pelvis showed splenomegaly, which measures 15 cm, and multiple collaterals in the splenic hilum region. A computed tomography angiogram [Fig- 4] demonstrated 3 fusiform aneurysmal dilatations noted along the splenic artery. The largest size, measuring 3.7×4.5 cm, was located in the lesser sac. One saccular aneurysm of size measuring 1.4×1.1 cm was present in the splenic hilum. She underwent laparotomy. Aneurysm excision with distal pancreatosplenectomy and esophagogastric devascularization was done [Fig-5 and Fig- 6]. During the postoperative period, the patient had a biochemical leak and was treated conservatively. During the follow-up, patient had normal blood counts and the upper gastrointestinal endoscopy revealed no esophageal or gastric varices.

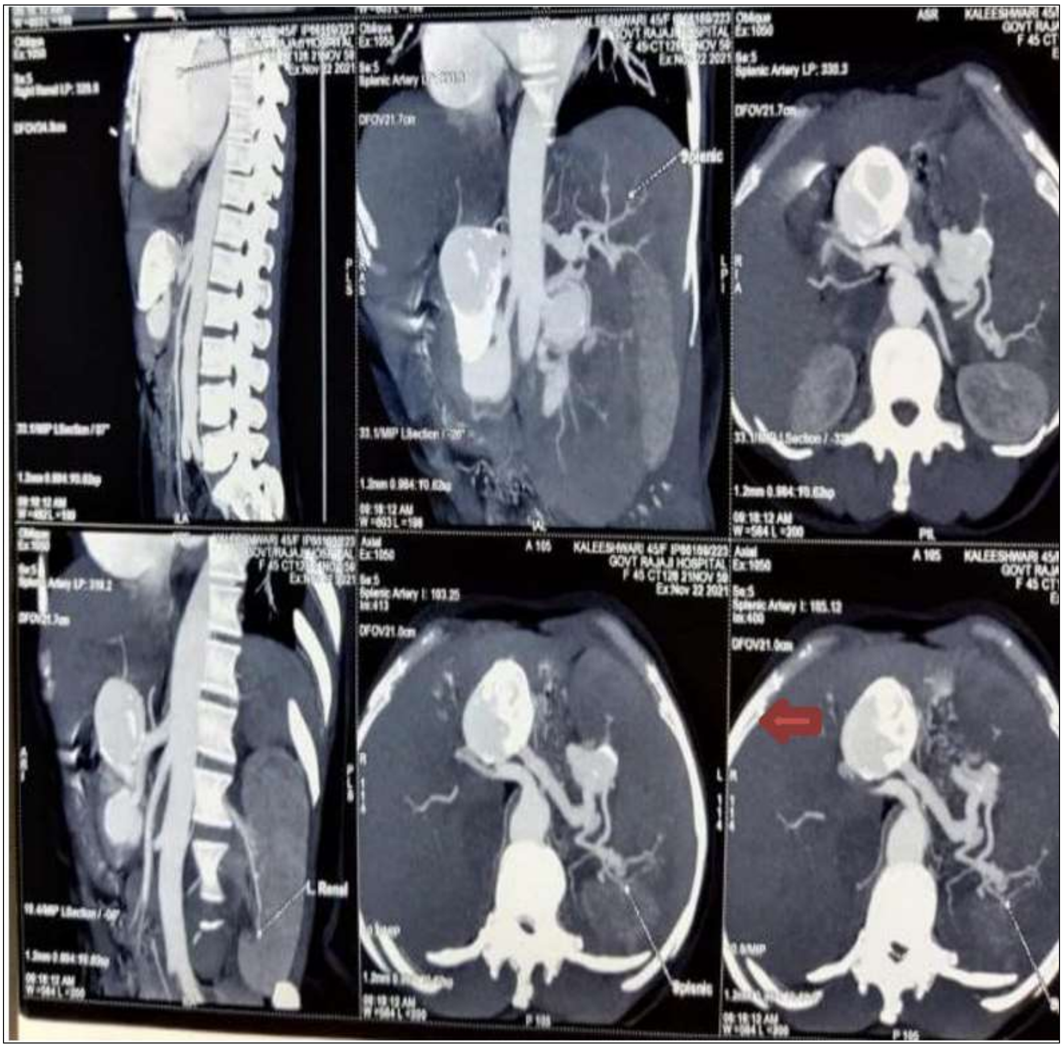

Fig 4: Computed tomography angiogram- Axial cuts showing splenic artey aneurysm with perisplenic collaterals (Arrow)

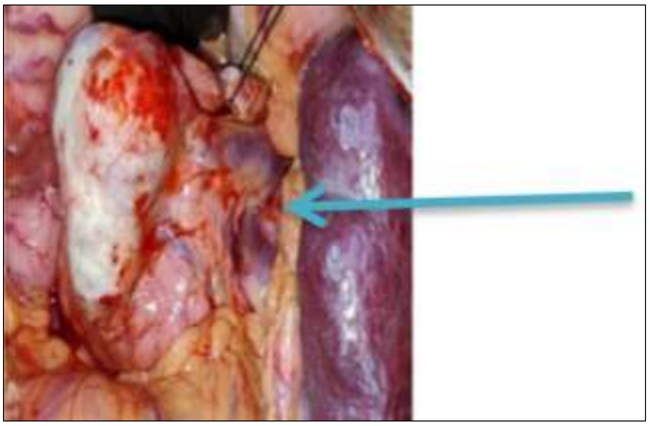

Fig 5: Intraoperative photograph showing Splenic artery Aneurysm with ligated splenic artery

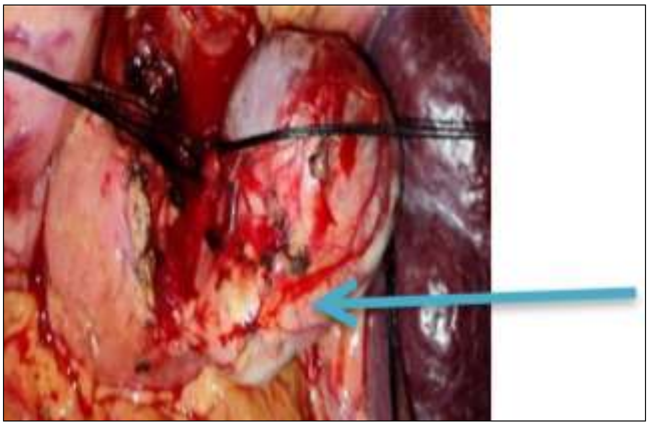

Fig 6: Intraoperative picture showing Aneurysm excision with Distal pancreatosplenectomy

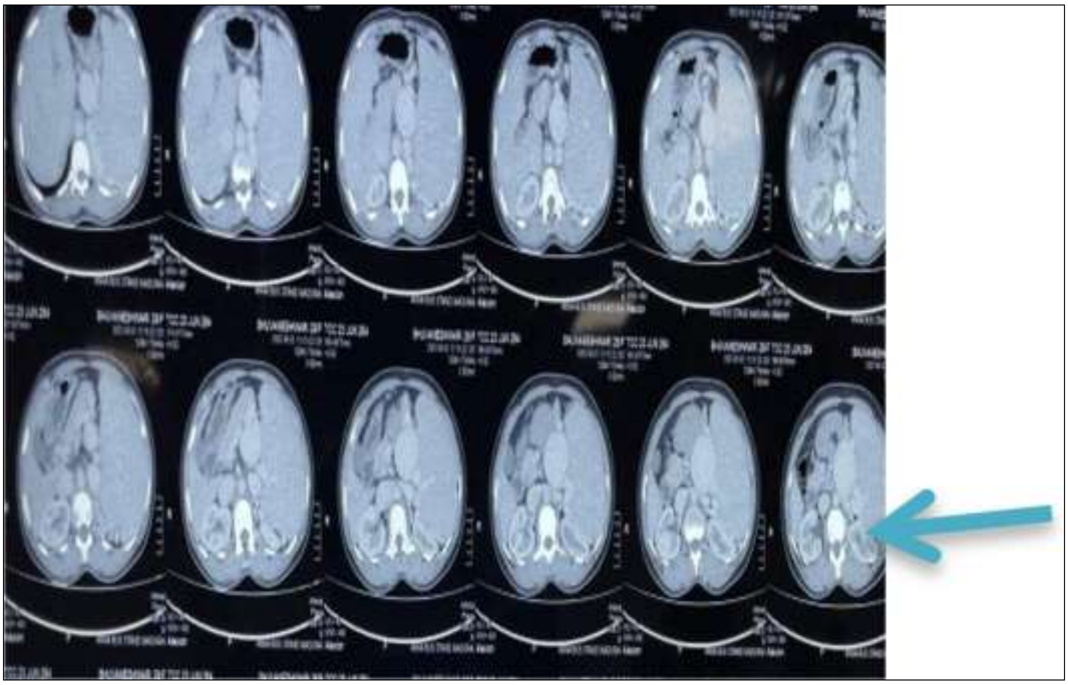

Fig 7: Contrast enhanced CT Abdomen & pelvis - Axial cuts showing Aneurysm in splenic artery with perisplenic , perigastric and peripancreatic collaterals

Case 3

A 28-years-old female patient presented with complaints of pain in the left hypochondrium and left lumbar region. Patient had a previous history of melena three years ago. The patient was evaluated and diagnosed as a case of Extrahepatic portal vein obstruction. Patients were managed conservatively. The upper gastrointestinal scope revealed Grade I esophageal varices with 3 columns and fundal varices. But patient was not started on any medication. On examination, the patient was pale. Moderate splenomegaly was present. The complete blood count results are Hemoglobin of 8.5gms/dl, a total count of 3000cells/mm^3, and platelet count of 85000cells/mm^3. Peripheral smear showed Pancytopenia. Bone marrow reported as reactive erythroid hyperplasia. Upper GI endoscopy findings are Grade I Esophageal varices with 2 columns and prominent fundal veins. Ultrasound of the Abdomen and pelvis with Doppler study revealed the main portal vein and its branches show chronic thrombosis with multiple dilated collaterals seen in the porta hepatis, peripancreatic and perisplenic region. Gross splenomegaly was present. A few well-defined aneurysms in the peripancreatic region, arising from splenic artery measuring 6x 5.4 cm and 4.8 x 3.8 cm each were present. Contrast-enhanced tomography of the abdomen and pelvis demonstrated bi-lobular fusiform aneurysmal dilatation of the distal part of the splenic artery measuring 5.6x 5.1x 6.7cm and 4.7x 3.9x4 cm, which displaces body and tail of pancreas anteriorly. Thrombus was noted in the main portal vein with adjacent periportal and perisplenic collaterals [Fig-7]. The patient underwent aneurysmal excision with distal pancreatosplenectomy and esophagogastric devascularization [Fig-8 and Fig- 9]. In the postoperative period, patient had a biochemical leak and was treated conservatively. During the follow-up, the patient had normal blood counts and the upper GI endoscopy revealed no esophageal or gastric varices.

Discussion

Splenic artery aneurysm is a rare clinical entity. The majority of patients are asymptomatic. The estimated prevalence rate of multiple splenic artery aneurysm is 0.02 to 0.1% [5].

Females are affected four times more commonly by SAA than men, especially during pregnancy and childbearing years due to endocrine changes**.** It is common in multiparous females due to high levels of oestrogen and progesterone resulting in the altered arterial wall and defective elastin synthesis, making the splenic artery more likely to dilate. Other risk factors include congenital anomaly of the vessels, vascular and connective tissue disorders, vascular trauma and portal hypertension [6].

The most common location of splenic artery aneurysm is at the mid or distal portion of the splenic artery, frequently found at the arterial bifurcation. In our patients there were multiple aneurysms located in the entire length of splenic artery in two cases and distal part of the splenic artery just proximal to the splenic hilum in one case.

The pathogenesis of EHPVO in SAA is due to compression on the splenic vein by the aneurysm leading to stasis of blood and thereby predisposing to thrombosis. Also, in portal hypertension, there exists a “splenic hyperkinetic state” which leads to increased splenic artery diameter and portosystemic shunts which reduce the resistance thus increasing the splenic vein flow. This hyperkinetic state promotes the development of SAA. Consequently, an increase in splenic artery blood flow is required to increase portal pressure and maintain liver perfusion resulting in the formation of a splenic artery aneurysm **[7].** This means either splenic arterial aneurysm can cause EHPVO or portal hypertension can cause splenic artery aneurysm.

The symptoms and signs of splenic artery aneurysm includes chronic abdominal pain, hematemesis, melena, acute rupture with hypotension, hemorrhagic shock and a sudden collapse which occurs in 3–10%. However, 80% of the patients found to be asymptomatic and are detected incidentally **[8].** In our series, the patients had symptoms of hematemesis, melena and abdominal pain with peripheral smear and ultrasound abdomen reports are pertaining to features suggestive of EHPVO. Multiple splenic artery aneurysms are diagnosed incidentally. The size of aneurysm in all three patients were more than 3 cm.

Treatment of SAA can be challenging and it is indicated in a pregnant woman with aneurysm, size less than 2 cms and in patients with portal hypertension, irrespective of size due to the high incidence of unpredictable rupture [9]**.** Treatment of SAA includes surgical or endovascular techniques. In patients with EHPVO and SAA, the treatment option depends upon the presence of bleeding from esophagogastric varices or portal biliopathy. The surgical procedures for EHPVO patient includes portosystemic shunts, esophagogastric devascularization, and the more recently described Rex shunt. The options for portosystemic shunts include proximal lienorenal shunt (LRS), side-to-side LRS, distal LRS, and mesocaval shunt. Surgery can simultaneously treat both problems in a single sitting. Endovascular therapy like coil embolization, balloon occlusion, and stent graft can be a good alternative in patients without bleeding or jaundice. The long-term complications of shunt surgery in EHPVO patients are shunt occlusion (5–10%) and recurrent bleeding. Post shunt encephalopathy is very rarely seen in EHPVO patients because of their preserved liver function.

All patients in our series underwent aneurysm excision with distal pancreatosplenectomy and esophagogastric devascularization. Post-operatively, two patients had biochemical leak (GRADE I POPF) which was managed conservatively. There was no mortality.

Conclusion

Multiple splenic artery aneurysm in the clinical background of EHPVO is very rare. Despite their rarity, it can be managed successfully by surgical intervention with good favourable outcomes.

Acknowledgement

Not available

Author's Contributions

Padmanabhan S proposed the study. Thamarai Kannan M and Karthikeyan S performed research and wrote the first draft. Thamarai Kannan M, Venkkatesh S, Krishna Prasad MB collected the data. Sastha A and Villalan R contributed to the design and interpretation of the study. Sastha A, Padmanabhan S, and Karthikeyan S contributed to further drafts.

Conflict of Interest

Not available

Financial Support

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