

Giant and Multiple Splenic Artery Aneurysms: Our Experience of Surgical Approach and a Brief Pathophysiological Review

Vijay Waman Dhakre^{1*}, Chetan Rathod², Kiron Krishnaprasad³ and Kayomarz Sohrab Sethna⁴

¹Assistant Professor, Department of General Surgery, Lokmanya Tilak Municipal Medical college and General Hospital, Sion, Mumbai, Maharashtra, India

²Associate Professor, Department of General Surgery, Lokmanya Tilak Municipal Medical college and General Hospital, Sion, Mumbai, Maharashtra, India

³Resident Doctor, Department of General Surgery, Lokmanya Tilak Municipal Medical college and General Hospital, Sion, Mumbai, Maharashtra, India

⁴Professor, Department of General Surgery, Lokmanya Tilak Municipal Medical college and General Hospital, Sion, Mumbai, Maharashtra, India

***Corresponding Author:** Vijay Waman Dhakre, Assistant Professor, Department of General Surgery, Lokmanya Tilak Municipal Medical college and General Hospital, Sion, Mumbai, Maharashtra, India.

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Abstract

A 39 year old gentleman with chronic liver disease due to HBV infection presented with upper abdominal symptoms. On investigating he was diagnosed to have Giant multiple true splenic artery aneurysms. Surgical approach was the treatment of choice and a total splenic artery aneurysmectomy was done. HBV infection can be associated with the formation of aneurysm. In this report we have shared our experience and reviewed the pathophysiology briefly.

Keywords: Splenic Artery Aneurysm; Chronic Liver Disease; HBV

Abbreviations

tSAA: True Splenic Artery Aneurysm; SA: Splenic Artery; CLD: Chronic Liver Disease; HBV: Hepatitis B Virus; PH: Portal Hypertension; PAN: Polyarteritis Nodosa

Introduction

True splenic artery aneurysms (tSAA) are rare entities in the general population. But when assessing upper abdominal pain in patients of portal hypertension (PH), it should be considered because of the high incidence in these patients. Endovascular intervention is the treatment of choice for high-risk tSAA but the presence of even rare scenario like multiple/Giant tSAA can make surgical intervention inevitable. In this case discussion, we tried to focus on this perplexing clinical situation.

Case Report

A thirty nine year old gentleman presented with continuous dull aching pain in epigastric region radiating to right hypochondriac region since 7 to 10 days. It was associated with anorexia and abdominal distension. There was no history of fever, constipation, melena or hematemesis. He has a past history of jaundice 20 years back which subsided without medication and was not investigated.

Clinically the patient was hemodynamically stable. Abdomen was uniformly distended, few visible distended veins over right and left upper quadrant of the abdomen. Umbilicus was normal and no evidence of spider nevi seen.

He had tenderness localised to right hypochondriac region on deep palpation. Murphy's sign was absent. Patient had ascites. Rest of the abdominal findings were normal.

There was no evidence of liver cell failure or pedal edema.

Investigation

His blood analysis showed Hb 11.3 g/dL and leucocytopenia (2900 per μ L) and thrombocytopenia (90000 per μ L), Liver function test was abnormal.

Total Bilirubin 1.8 mg /dl, Direct Bilirubin 0.5 mg/dl, SGOT 142 IU/L SGPT 120 IU/L.

Alkaline phosphatase 94 IU/L, Total protein 5.2 g/dL and serum albumin 3.2 g/dL with normal INR of 1.2. Serum sodium was 136 mmol/L and rest of Renal function test was normal. Serum Lipase 20 units/L.

Abdominal ultrasound was done which showed Nodular liver with features of portal hypertension in the form of loss of portal vein phasicity and dampened flow to portal vein with multiple portosystemic collaterals and multiple splenic artery aneurysms largest 6.3 x 5.0 cm near its origin from the celiac axis.

Diagnostic ascites tapping showed a high Serum albumin ascites gradient (SAAG) 2.7 without any granulocytic infiltration.

An abdominal computed tomography (CT) scan (Figure 1 and Figure 2) was done to evaluate this aneurysm which suggested a dilated tortuous splenic artery (SA) with multiple aneurysms as follows:

1. 6.0 x 4.3 x 5.3 cm wide neck saccular aneurysm from the proximal retropancreatic portion around 2.7 cm from its origin, causing mass effect on the proximal pancreas and having an anterior out-pouching suggestive of impending rupture.
2. 4.1 x 2.7 x 3.5 cm sized fusiform aneurysm arising from the mid part of splenic artery at the level of left renal vein.
3. 3.7 x 3 x 3.7 cm saccular aneurysm from the distal splenic artery closely abutting the body of stomach with a focal thrombus in the superolateral lobulated part.
4. 4.6 x 2.4 x 3 cm trilobed aneurysm in splenic hilum with multiple calcifications within.

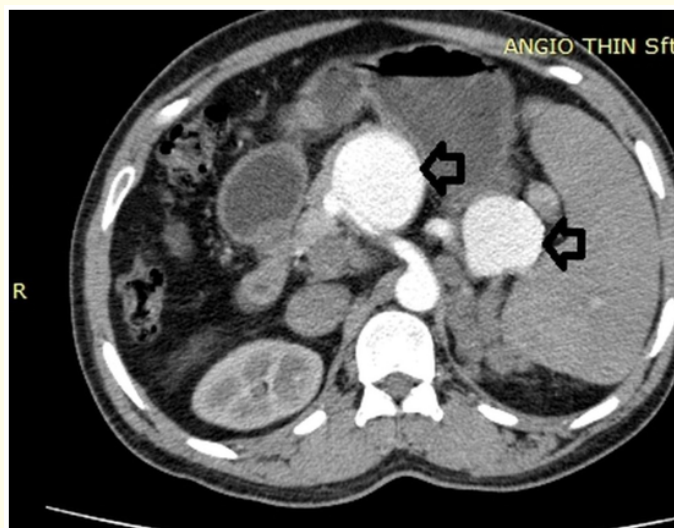


Figure 1: Transverse view of Giant splenic artery aneurysms (black arrows) arising near the origin and near the hilum of spleen.

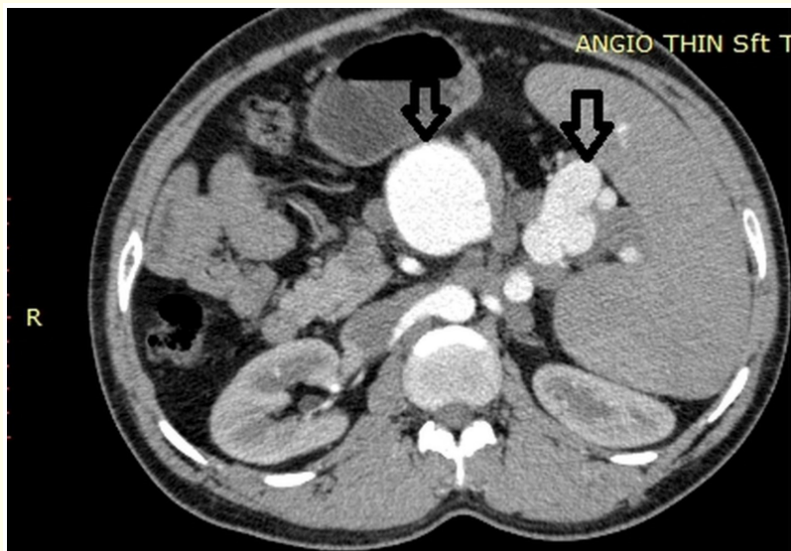


Figure 2: Transverse view of Giant splenic artery aneurysms (black arrows) arising near the origin and middle part of splenic artery.

All these above features suggested that the aneurysms were true aneurysms (no evidence of inflammation or infection) and also showed caudate lobe hypertrophy, ascites and mild splenomegaly. The etiological workup for chronic liver disease was done in which he was found to be Hepatitis B surface antigen positive. HBV DNA viral load was 141 IU/ml and HBeAg was negative. Hence, he was diagnosed to have decompensated chronic liver disease.

Upper GI endoscopy showed one large varix and one small varix in oesophagus for which endoscopic variceal banding was done.

The patient was thus optimized for surgery with a CTP 9/B of and MELD-Na 14 and was started on Tablet Entecavir 0.5 mg once a day.

A bilateral subcostal incision was made which showed 4 large aneurysms identical to what was described in the CT angiography was noted.

SA was approached from the lesser sac and traced which lead us to the most proximal aneurysm located around 1 cm from the celiac axis over the SA (Figure 3). Three more such but comparatively smaller pulsating aneurysms were noted along the superior border of the pancreas.

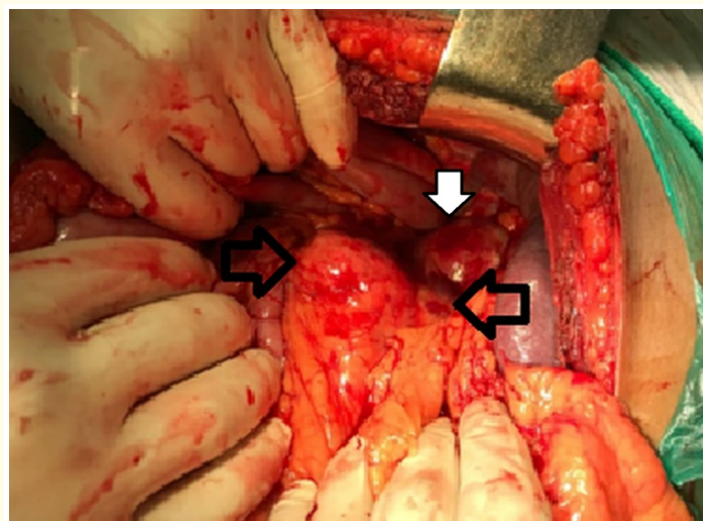


Figure 3: Intra-operative view showing tSAA near the origin and middle part of SA (black arrows) and near the splenic hilum (white arrow).

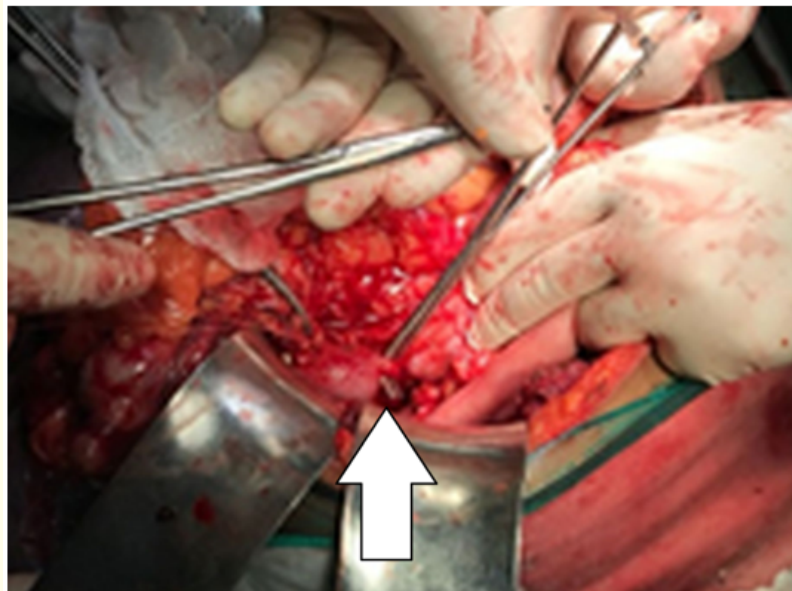


Figure 4: Intra-operative view showing dissection and looping of SA origin (white arrow) note the collapsed aneurysm near the origin of SA.

The SA was ligated and following which the aneurysms collapsed and became soft but within few seconds the aneurysms filled up and started pulsating (Figure 4) like before thus suggesting patent collaterals from Inferior pancreatic and left gastroepiploic arcades. Due to this 'refilling aneurysms' a complete splenic artery excision with splenectomy was performed.

Patient was extubated in the theatre and observed in ICU for the next 24 hours. He subsequently became stable enough to get transferred toward and had a hospital stay of 7 days with grade 2 Clavien-Dindo Classification for postoperative complication.

Discussion

True splenic artery aneurysms (tSAA) are very rare entities. They are mostly solitary but multiple tSAA can be encountered. The splenic artery is one of the commonest arteries to undergo aneurysms in the splanchnic arterial system. It is prone to both pseudo- and true aneurysms. We will restrict our discussion to true aneurysms only. The true aneurysms are mostly related to congenital causes (viz congenital defects of the splanchnic artery) or acquired (viz. arterial fibrodysplasia, atherosclerosis, trauma, pregnancy) [1,2].

In majority of settings the tSAA goes unnoticed and is incidentally found during abdominal radiological scans or during the autopsy, the incidence can range from 0.01% in general population to 10.4% in selected groups [1,2].

We want to focus on the fact that there is a specific group of patients with portal hypertension (PH). The incidence in this group may vary from 7.1% to 20% [2-5].

3% to 10% of patients present with haemorrhagic shock. Acute presentation with epigastric pain with positive Kehr's sign may indicate ruptured splenic artery aneurysm. Patients at high risk of rupture are pregnancy, portal hypertension, Caroli's disease or patients who have undergone Portocaval shunts or after liver transplant. Any aneurysm exceeding 2 cm in diameter is at risk for rupture [1,2].

Only 20% of these aneurysms are symptomatic. Due to a high incidence in PH patients, it needs to be included in the differential diagnosis of epigastric pain with or without left hypochondrial pain [2]. Systemic symptoms like anorexia, nausea or vomiting can be associated which can create confusion and delay in diagnosing tSAA.

Our patient had abdominal distension and epigastric and right hypochondriac pain. He also had decompensated CLD due to hepatitis B virus with portal hypertension.

tSAA and portal hypertension

In PH there are major shifts in portal blood pressures and along with abnormalities in splanchnic blood flow. This can be picked up by calculating a ratio of splenic and hepatic artery proper diameters. This incidence of complications associated with PH is found to be 83.4% if this ratio is more than 1.4 [4]. Increased portal blood flow in portal hypertension is postulated to be one of the leading cause of tSAA. There is reactionary increase in splenic artery (SA) flow in response to the portosystemic shunting. The resultant increase in splenic artery flow will predispose the SA to form tSAA.

tSAA associated with pregnancy and metabolic changes

tSAA and its rupture is strongly associated with pregnancy. Increase estrogen-progesterone levels during pregnancy alter receptors on the artery causing aneurysmal changes. Another hormone affecting the integrity of arterial wall is hormone relaxin which is secreted in later part of pregnancy. Also, there is increased porto-venous congestion seen in pregnancy which can indirectly cause increase SA blood flow and can precipitate tSAA [8].

Changes in collagen metabolism due to cirrhosis increases the risk of aneurysm formation and rupture.

These metabolic changes like the collagen lysis predominantly seen in patients with cirrhosis with PH, post laparotomy and post liver transplant makes this group of patients as high risk group for tSAA formation and rupture.

tSAA association with HBsAg, cryoglobulinemia and Polyarteritis Nodosa (PAN)

tSAA was found to be associated with viral hepatitis (mostly hepatitis B virus (HBV) as in our case) in about 15.4% to 17% of cases [3].

PAN is associated with HbsAg positive patients and is shown to induce vasculitis that resembles PAN excluding orchitis. This association is postulated to be due to immune complex deposition in major vessels or due to excess circulating antigen load.

Cryoglobulinemia is seen in 0-15% of HBV infected patients. Also, precipitation of immunoglobulins is observed in a cold environment in HBV infection which is reversible. This can cause cryoglobulinemia-induced vasculitis which can potentially give rise to the aneurysm. But major artery aneurysm is more associated with PAN.

All tSAA don't warrant active treatment. Treatment is offered to aneurysms which are more than 2 cm and found to be increasing in size on surveillance. Intervention is also considered in cases associated with portal hypertension and in tSAA with pregnancy [2,7]. The surgical approach is the best approach for multiple and giant tSAA. The tortuous course of SA and multiple tSAA makes the endovascular approach less feasible [2,7,8]. Open surgical approach with aneurysmectomy with or without splenectomy is well described in the literature. The only ligation in continuity can cause refiling of aneurysms hence complete excision is sometimes inevitable [2,3,7,8].

Conclusion

Multiple tSAA are very rare and can have complex pathophysiology. tSAA can be a complication of HBV infection and should be included as a differential diagnosis while assessing a tender abdomen especially with portal hypertension. Complete excision of SA with splenectomy is sometimes inevitable and most feasible approach.

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