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Ligation of symptomatic celiac artery aneurysm without vascular reconstruction: Utilizing the natural collateral circulation of the celiac axis: A case report

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ABSTRACT

INTRODUCTION: Celiac artery aneurysm is very rare visceral artery aneurysm. Symptomatic and ≥ 2.5 cm sized aneurysm requires treatment. Excision and revascularization is the most commonly employed procedure.

CASE PRESENTATION: We report a case of ligation and excision of celiac artery aneurysm extending onto the splenic and hepatic arteries without vascular reconstruction. The patient was a 52 year old lady who was evaluated for abdominal pain and was found to have a celiac artery aneurysm involving the hepatic and splenic arteries. She was evaluated with computerized tomography and digital subtraction angiography of the abdominal vessels. These confirmed good natural collaterals from the branches of superior mesenteric artery supplying the liver, stomach and spleen. We performed ligation and excision of the aneurysm and ligation and division of hepatic, splenic and left gastric arteries as the aneurysm was extending on to these vessels, without any vascular reconstruction, utilizing the natural collaterals from the superior mesenteric artery.

DISCUSSION: Ligation of celiac artery aneurysm without revascularization is often done in emergency situations. Excision and revascularization is the treatment of choice to ensure adequate blood supply to liver, spleen and stomach. We could utilize the natural collateral circulation of celiac artery from superior mesenteric artery avoiding a complex procedure of revascularization.

CONCLUSION: We present this because of the rarity of the disease as well as rarity of the technique of not performing vascular reconstruction. We emphasize on the pre-operative and operative evaluation of collateral circulation with conventional angiography and intraoperative Doppler respectively.

1. Introduction

Celiac Artery Aneurysm (CAA) is a very rare visceral artery aneurysm (VAA). It accounts for 4%–30% of all the visceral artery aneurysms [1,2]. The common causes of CAA in the contemporary era are congenital medial defect or atherosclerosis [3]. Though majority of these are asymptomatic and are picked up by cross sectional imaging, treatment is warranted in CAA >2 cm in diameter, because of the higher risk of spontaneous rupture [4]. Rupture of CAA is a catastrophic complication with mortality rate of 40%, even with surgical intervention [5]. Isolated CAA is even rarer. More commonly it is associated with other VAAs [6,7]. Treatment options depends on the size, associated other VAAs, concomitant aneurysmal involvement of hepatic, splenic and left gastric arteries as well as the presence of collaterals. Surgical treatment in the form of resection or ligation of the aneurysm with vascular reconstruction of the celiac trunk to the hepatic and splenic arteries is the preferred choice [6,8,9]. Resection or ligation without reconstruction is usually not recommended because of the possibility of hepatic, gastric and splenic ischemia [10]. In this case report we present a case which was managed in our private tertiary care, multispecialty institution by ligation of CAA involving the hepatic and splenic arteries without any vascular reconstruction emphasizing the utilization of natural collaterals of celiac axis from superior mesenteric artery. The objective of presenting this case is to bring to light the possibility of utilizing the existing collateral circulation

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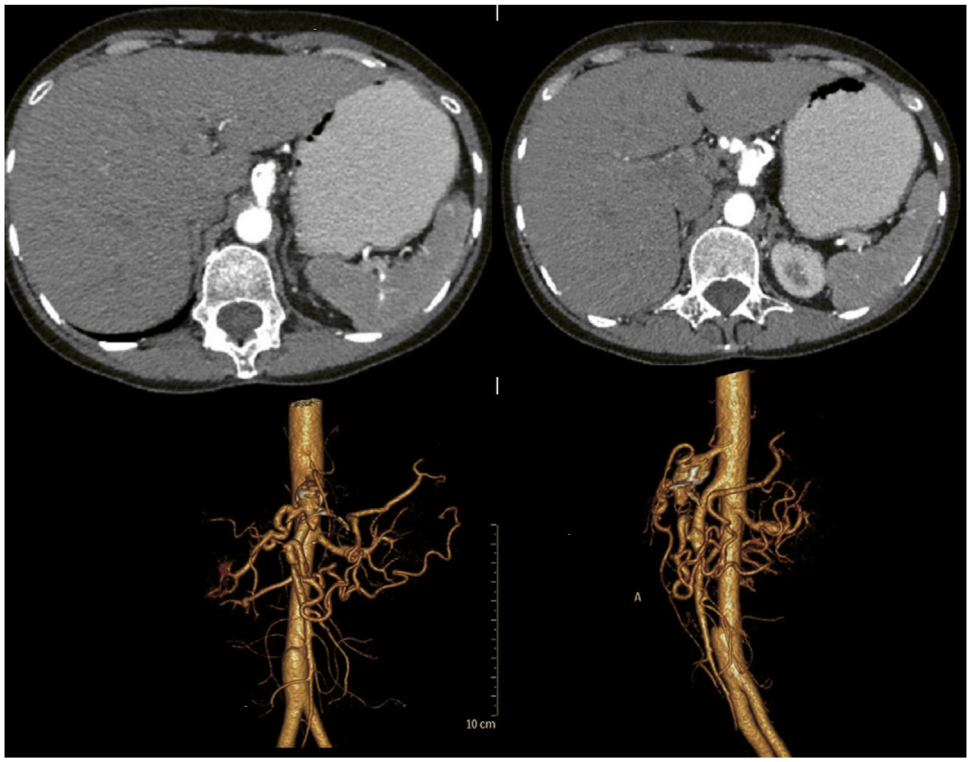

Fig. 1. Computerised angiography showing CAA involving the splenic and hepatic arteries, with partially occluding thrombus.

of the celiac artery and thus avoiding more complex resections and vascular reconstruction in selected patients with CAA.

2. Case presentation

This was a 52 year old lady who presented as an outpatient to us in August 2018 with on and off epigastric pain radiating to the back. She was diagnosed to have CAA in October 2017, for which she was on follow up. She had remained symptomless until the present outpatient visit. She does not have any relevant significant past or pharmacologic history. Ultrasonogram of the abdomen revealed a 2.8 cm aneurysm of the celiac artery with extension of aneurysmal dilatation onto the hepatic and splenic arteries. For further evaluation, Computerized Tomographic (CT) visceral angiography was done. This confirmed the ultrasound findings as well as detected partially occluding thrombus in the celiac and splenic artery aneurysms (Fig. 1). Smaller aneurysms were found in the Superior Mesenteric Artery (SMA) and both renal arteries.

Conventional Digital Subtraction Angiography (DSA) was done in February 2019 to assess the collateralization and to further delineate the vascular anatomy. This showed a replaced right hepatic artery, prominent inferior pancreaticoduodenal and gastroduodenal arteries in addition to new collateral formation around the splenic artery (Fig. 2). Although DSA had shown adequate collateral flow, she was planned for open ligation, excision with revascularization of the branches of celiac trunk. This was because all the 3 branches of the celiac trunk were aneurysmally dilated for up to 2 cm from their origins from the celiac artery. We were unsure whether we could successfully retain adequate collateral circulation. Endovascular therapy (EVT) was not considered as the aneurysm was very close to the origin of the celiac artery from the aorta. Abdomen was explored through a midline laparotomy. This showed 2.5 cm diameter aneurysmal dilatation of the Celiac artery less than 0.5 cm from the point of its origin, extending to the hepatic, left gastric and splenic arteries. The wall of the aneurysm was heavily calcified. After isolating the supra celiac aorta, the origin of the celiac artery was dissected and encircled. The hepatic, splenic and left gastric arteries were also individually dissected and encircled with vascular loops. The supra celiac aorta was not clamped at all during the procedure, thus avoiding the risks of ischemia. The root of the celiac artery at the origin was occluded with vascular clamp to assess the blood flow to the liver, stomach and spleen. After confirmation of blood flow to the liver by palpation of pulsation and intraoperative doppler, the hepatic artery and the splenic arteries were also occluded at a point just distal to the aneurysm. Once normal vascularity of the liver, stomach and spleen was confirmed by visual inspection, palpation and doppler (for hepatic arterial flow), the decision for revascularization was revised. The celiac artery was suture ligated at the origin, the hepatic and splenic arteries were also suture ligated and divided at the post aneurysmal area and the celiac artery aneurysm was opened (Fig. 3). Thrombus was removed, aneurysm partially excised and the stumps on both ends were over sewn with 3-0 polypropylene sutures. The procedure was performed by a team comprising of Cardiovascular and thoracic surgeon with 20 years of experience in aneurysm surgeries and Hepatopancreatobilary (HPB) surgeon with 20 years experience in HPB surgery.

She had an uneventful post-operative recovery. She continues to be asymptomatic at 1 year follow up. A repeat CT angiography of the abdomen was done 6 months after the surgery. This has shown normal arterial supply of the liver, stomach and spleen (Fig. 4).

This work has been reported in line with SCARE 2018 criteria [11].

3. Discussion

Surgical excision/ligation and revascularization is the preferred treatment, in most centers, for CAA $\geq$ 2.5 cm diameter, even in asymptomatic patients who are good surgical candidates [6,10,12]. Ligation without reconstruction is commonly performed in case of

Fig. 2. Digital Subtraction Angiography shows the CAA and associated extensive collateral blood vessels from the splenic artery and around the stomach.

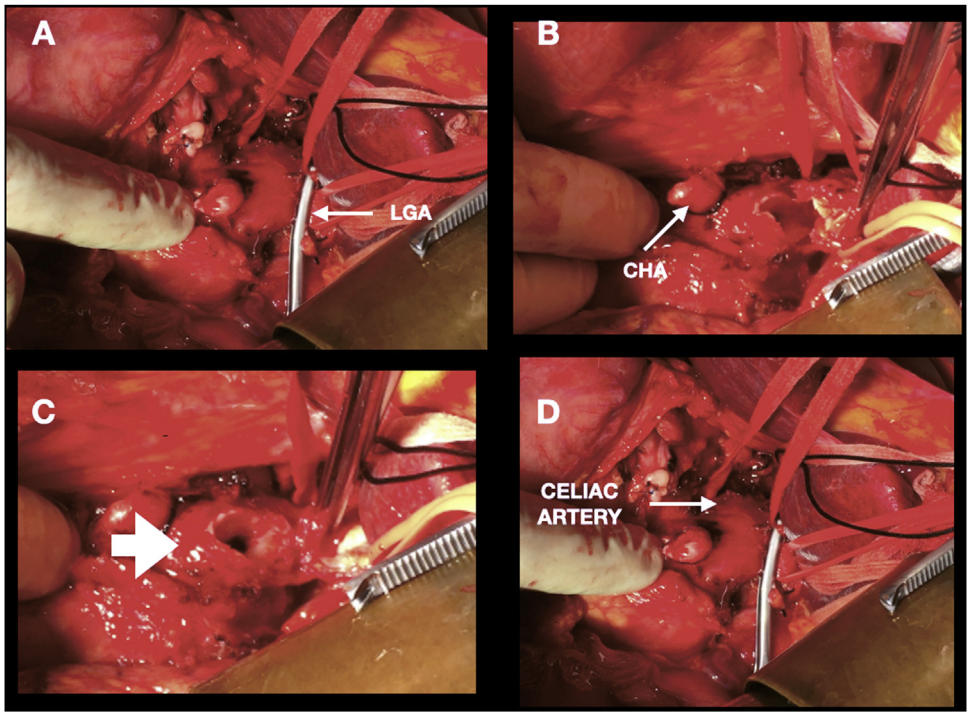

Fig. 3. LGA Left Gastric Artery, CHA Suture ligated stump of Common Hepatic Artery, Thick arrow shows the opened aneurysm.

emergent situations like spontaneous rupture. Ligation and excision can be considered in elective situations only after confirmation of adequate collateral circulation from the branches of Superior Mesenteric Artery (SMA) [13]. However, there have been very few reports of surgical treatment of CAA without revascularization in elective situations. In one of the previous case reports of excision of CAA, distal pancreatosplenectomy, had to be performed because of the presence of severe peri-lesional adhesions making it impossible to separate the aneurysm from the superior border of pancreas [14]. The authors had not considered ligation alone and had not described the any pre-operative evaluation for natural collateral circulation of celiac artery. This kind of operation is of significant magnitude and is prone for major complications. In another series of 9 elective surgical interventions for CAA, one patient underwent ligation without revascularization [6]. The authors have not mentioned the reason for opting such an approach. Vascular reconstruction makes the procedure more technically challenging and puts the patient in need for, at least short term anticoagulant therapy, though long term therapy is often not necessary. EVT is yet another option for VAA which has given good long term success. There are only a few case reports on EVT for CAA and data on long term outcomes is still evasive [15,16]. EVT is effective in high surgical risk patients and in those who had an abdominal surgery in the past. This is usually a combination of different modalities like occluding the distal arteries and coiling the aneurysm, injection of embolic material, deployment of covered stent or flow diverting stent and percutaneous thrombin injection. However, endovascular stenting was not considered in our patient as the aneurysm was arising very close to the origin of the celiac artery. Coiling was deemed unsafe due to the anatomy of the aneurysm and there was serious concern of end organ ischemia as well as the anatomy of the aneurysm involving hepatic, left gastric and splenic arteries.

Thus, our proposition from this case report is that collateral formation of the vasculature of the organs supplied by the celiac artery should be assessed before and during the procedure with DSA and temporarily occluding the branches of celiac artery respectively.

Fig. 4. Computerised Tomography of the abdomen performed 6 months post operatively showing good perfusion of the liver, stomach and spleen through the naturally formed collaterals.

We could avoid vascular reconstruction by utilizing the natural collaterals of the celiac axis from the SMA. The right lobe of the liver was adequately perfused by the right hepatic artery. The natural collateral circulation from the SMA to the proper hepatic artery was maintained through the gastroduodenal artery. The stomach was perfused by the retrograde flow from GDA through the right gastroepiploic and right gastric arteries. In addition there were new collaterals around the splenic artery which were perfusing the spleen (Fig. 5).

CAA being a rare disease and as reports on excision avoiding vascular reconstruction are even rarer, our approach cannot be recommended as a gold standard approach to management of CAA. This case report emphasizes the utilization of natural collaterals of celiac artery in patients undergoing surgery for CAA which helps avoiding complex vascular reconstructions. This can be safely performed in selected patients after careful pre-operative and intra-operative assessment of collateral circulation thus bringing down the complexity and morbidity of the procedure.

Declaration of Competing Interest

The authors report no declarations of interest.

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Ethical approval

It was exempted from ethical approval as this is a case report.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Concept/Design: Deepak Varma, Binoy Chattuparambil **Analysis/interpretation:** Dhruvakuma Krishna **Data Curation**: Sripadh Upadhya, Sreedhar Devaraju **Writing draft manuscript:** Deepak Varma, Sreedhar Devaraju **Critical revision:** Sripadh Upadhya, Dhruvakumar Krishnan **Approval of manuscript:** Binoy Chattuparambil, Dhruvakumar Krishnan **Agreement to be accountable:** Deepak Varma, Binoy Chattuparambil, Sripadh Upadhya, Sreedhar Devaraju and Dhruvakumar Krishnan.

Registration of research studies

Not applicable.

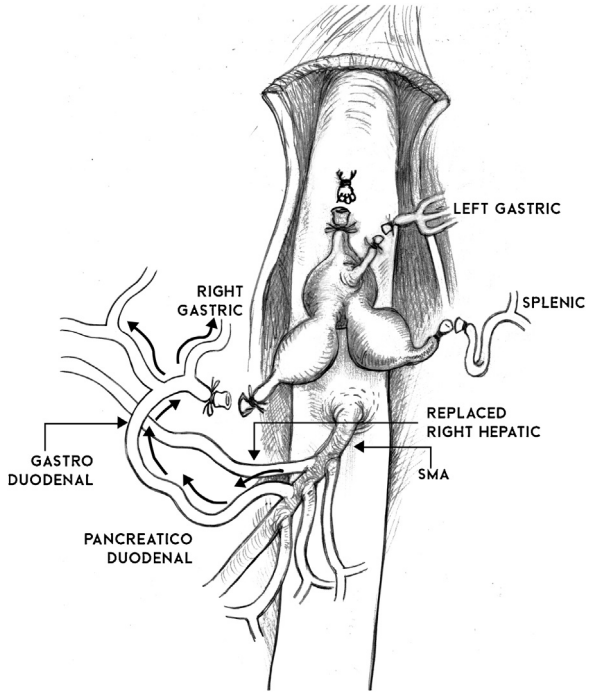

Fig. 5. Diagrammatic representation of the collateral circulation after severing the arteries involved by the aneurysm in our patient.

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