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Aneurysm of anomalous splenic artery arising from a splenomesenteric trunk: Review of the literature with a report of a new case

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ABSTRACT

INTRODUCTION: Aneurysm of splenic artery arising from splenomesenteric trunk is an extremely rare condition. The aim of this study is to report a new case with literature review.

PRESENTATION OF CASE: A 52-year-old housewife presented with mild central abdominal pain for two month duration. Abdomen was soft. Abdominal ultrasound examination showed a focal aneurysmal dilatation in the splenic artery (SA) near the portal vein. Abdominal computed tomographic angiography (CTA) revealed presence of the splenomesenteric trunk with fusiform aneurysm (45 × 33 mm) of the proximal part of the SA. In supine position, through upper midline laparotomy incision, exploration of both superior mesenteric artery (SMA) and SA was performed, total excision of the aneurysm was done, the SMA was side-repaired and SA was ligated. The post-operative period was uneventful.

DISCUSSION: It is interesting to note that orthotopic SA aneurysms, most commonly present in the distal third of the artery, followed by the middle third, while in cases of splenomesenteric trunk, all reported cases of anomalous SA aneurysms including the current one, showed the aneurysms to be located in the proximal portion or root of the SA.

CONCLUSION: Splenomesenteric trunk is a rare anatomical anomaly, aneurysm of which is even rarer. It can be managed either by endovascular intervention or open surgery.

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1. Introduction

Visceral artery aneurysm represents a dangerous vascular disease that can potentially be fatal, albeit it is a rare condition [1].

Splenomesenteric trunk is an anomalous SA that anatomically originates from SMA with less than 1% of the population having this variant. Aneurysm of this rare anatomical variant is even rarer with occasional case reports in the English medical literature, since SA aneurysm is a rare condition in itself, never the less in an anomalous

SA. The reports are spread across 29 English medical literatures with the first report been published in 1967 by Ghatan et al. [1].

Though extremely uncommon, they still have clinical importance as the therapeutic strategies for an anomalous SA aneurysm may significantly differ from those of orthotopic anatomical origins and their rupture may have disastrous consequences.

The aim of this study is to report a case of aneurysm of anomalous SA arising from a splenomesenteric trunk with a brief review of the literature. The report has been arranged in line with SCARE 2018 guidelines [2].

1.1. Patient information

A 52-year-old housewife presented to outpatient clinic with mild central abdominal pain for two month duration. The patient

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Fig. 1. 3D reconstruction CT angiography shows splenomesenteric trunk with fusiform aneurysm involving the proximal part of the splenic artery.



Fig. 2. Intraoperative finding of the aneurysm.

had history of open cholecystectomy before 20 years with a negative past medical history. No family history was reported for the same complaint. The patient denied drug allergy.

1.2. Clinical findings

The patient was stable (blood pressure: 130/75 mmHg, pulse rate: 88 beats/minute, respiratory rate 13 cycles/minute, temperature 37.2C), there was a right subcostal incision scar. Abdomen was soft, no mass was felt. Pancreatitis, chronic appendicitis, gastric ulcer, duodenal ulcer and visceral vessel problems were suspected.

1.3. Diagnostic assessment

Hematological tests were normal. Abdominal ultrasound examination showed a focal aneurysmal dilatation seen in the splenic artery near the portal vein. Abdominal CTA revealed presence of the splenomesenteric trunk with fusiform aneurysm (45 × 33 mm) of the proximal part of the SA (Fig. 1).

1.4. Therapeutic intervention

The patient received meningococcal conjugate vaccine two weeks before the intervention. The patient was prepared for general anesthesia, she was injected a vial of ceftriaxone (1 g IV) one hour before the intervention, in supine position, through upper midline laparotomy incision, exploration of both SMA and SA was performed (Fig. 2), total excision of the aneurysm was done, the SMA was side-repaired and SA was ligated. The laparotomy wound

Table 1
Socio-demographic and clinical characteristics of the reported cases of the splenic artery aneurysm arising from the splenomesenteric trunk.

Variables	Total case, n = 45
Age (years)	46 (22–73)
Gender	
Female	26 (57.8 %)
Male	19 (42.2 %)
Ethnicity	
Asian	26 (57.8 %)
European	14 (31.1 %)
American	5 (11.1 %)
Symptoms	
Asymptomatic	26 (57.8 %)
Abdominal pain	19 (42.2 %)
Aneurysm size (largest diameter in cm)	2.95 (2–6)
Treatment	
Surgical resection	18 (40 %)
Coil embolization	12 (26.7 %)
Coil embolization + laparoscopic ligation of the splenic artery	2 (4.4 %)
Graft placement	3 (6.7 %)
Stent-graft placement + coil embolization	7 (15.6 %)
None	3 (6.7 %)
Splenectomy	4 (8.9 %)
Mortality	None is reported

was closed in classical way. The operation was performed by the first, second, sixth, eleventh and twelfth authors. They have eight year experience in fields of vascular and general surgery.

1.5. Follow-up and outcomes

The post-operative period was uneventful, the patient received IV Paracetol bottle 600 mg x3 with Tramadol ampoule on need. She was discharged home on the third postoperative day. The patient was followed up every one month for the first 4 months by phone call.

2. Discussion

During embryonic development, the primitive SAs may have an unusual variation allowing for SA to originate from SMA in less than 1% of population [3,4]. The incidence of SA aneurysm is estimated to be around 0.8 %, though the exact number is not known [1]. We searched through the Medline, PubMed, and Google scholar and were able to find only 44 reported of anomalous SA aneurysms (Table 1) [1,5–30]. The combined rarity of these two conditions explains why so few cases are reported in English medical literature.

It is interesting to note that orthotopic SA aneurysms most commonly present in the distal third of the artery, followed by the middle third, while in cases of splenomesenteric trunks, all reported cases of anomalous SA aneurysms including the current one, showed the aneurysms to be located in the proximal portion or root of the SA, behind the pancreas as it originates from SMA [31]. The exact cause for this is yet to be known.

Splenic artery aneurysms are often associated with atherosclerosis, pancreatitis, essential hypertension, portal hypertension, pregnancy and others in the literature. They are often found incidentally as most are asymptomatic [1]. In this case, the precise risk factor could not be found.

The anomalous SA arises at a right angle from SMA, usually from the its proximal part and courses upward a short distance. This large angle might lead to sudden hemodynamic changes with subsequent elevated tension at the vessel wall and its derangement, we speculate that this is the cause of the aneurysm at the proximal portion of the anomalous artery repeatedly.

Initially Ultrasound imaging can be used for the diagnosis, then confirmed by CT angiography and to further evaluate the size of the lesion and the morphology of the involved arteries, as the aorta and its branches can be shown in constructed 3D images. Invasive catheter-based angiographies are no longer routinely used due to availability of CTA unless an endovascular procedure is planned for treatment. Generally, angiography provides valuable information for planning the treatment strategy, such as the adequacy of collateral circulation and the choice to preserve the spleen. In the current case, multiple collaterals could be visualized on CT angiography that is why the spleen was left in place [1,13].

The methods used in treatment of splenic artery aneurysm have been used in treatment of anomalous SA aneurysms with good results and they include open or laparoscopic surgical repairs as well as endoscopic approaches [32].

In the reported case series, eighteen of them were treated by open surgery. The operation for an anomalous SA aneurysm is technically demanding and exposure of the aneurysm due to its location leads to greater surgical trauma. The procedure entails resection of the aneurysm with/without splenectomy and/or distal pancreatectomy.

Since 2005, a trend can be seen in the literature where endovascular treatments are increasingly being favored over open surgery. The endovascular approach entails several options. Treatment can be done by stent-graft implantation or by the isolation technique, where the inflow and outflow arteries are occluded or by embolization of the aneurysm itself, a procedure called packing technique. When dealing with splenomesenteric trunk aneurysm, a stricter criterion is needed for the selection of cases for endovascular treatment compared to the normal origin, since there is the risk of coil migration into the SMA with subsequent ischemia of the intestine. The neck of the aneurysm should allow the aneurysmal sac to be embolized properly with reduced distal migration risk. [13] In this case, endovascular intervention was not even an option because of lack of facilities.

The risk of splenic infarction from occlusion of SA is reduced by the presence of collateral arteries from left gastric or gastroepiploic arteries. None the less most splenic infarctions are well tolerated by the patient and are asymptomatic without sequelae or need for further management, making the presence of collateral circulation not indispensable [33].

Conclusion: splenomesenteric trunk is a rare anatomical anomaly, aneurysm of which is even rarer. It can be managed either by endovascular intervention or open surgery without splenectomy.

Declaration of Competing Interest

The authors report no declarations of interest.

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Ethical approval

Approval is not necessary for case report in our locality.

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.

Authors contribution

Fahmi H. Kakamad: Surgeon performing the operation, follow up the patient, writing the manuscript and final approval of the manuscript.

Zuhair D. Hammood, Bzhwen Y. Abdalla, Sanaa O. Karim, Hawar A. Sofi Mohammed, Shakhawan I. Hussein, Sana B. Anwar, Usama Y. Abulkarim: assisting in the operation, final approval of the manuscript.

Abdulwahid M. Salih, Karzan Mohammed, Hussein M. Hamasalih, Rawezh Q. Salih, Shvan H. Mohammed, Othman O. Qadr, Hawar A. Sofi Mohammed: major contribution to the idea, revision and final revision of the manuscript.

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