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Case Report

Endovascular Treatment of a Saccular Aneurysm in the Celiomesenteric Trunk: A Case Report and Review of Literature

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To present a world-first case of a successful endovascular treatment of a celiomesenteric trunk (CMT) aneurysm. A 45-year-old man had an asymptomatic saccular aneurysm in a rare anomaly of CMT. Endovascular multiple micro-coil embolization of the common hepatic artery, splenic artery and the aneurysm was done, followed by a stent-graft deployment in the superior mesenteric artery covering the orifice to the aneurysm. Postoperative course was uneventful. Only 21 cases have been previously reported in the literature, and all were treated by open surgeries. Endovascular therapy can be safely done in selected cases of a CMT aneurysm with sufficient collaterals to the liver and spleen.

Key Words: Aneurysm, Anomaly, Celiomesenteric trunk, Coil embolization, Stent-graft

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INTRODUCTION

The celiomesenteric trunk (CMT) is a rare developmental vascular anomaly sharing common origin of the celiac trunk and superior mesenteric artery (SMA) from the aorta [1]. An aneurysm in the CMT is very rare, but a challenging disease to treat because of the complex anatomic location. To our knowledge, this is the first case ever reported on a successful endovascular treatment of a CMT aneurysm.

CASE

A 45-year-old man was referred to our vascular clinic for a CMT aneurysm, which was incidentally found by computed tomography (CT) during a routine check. He was otherwise healthy without any symptoms. The diameter of the saccular aneurysm was 21 mm and located in the celiac trunk between the SMA orifice and celiac bifurcation (Fig. 1A). The gastroduodenal artery (GDA) was patent to form a good collateral circulation between the SMA and common hepatic artery (CHA). The left gastric artery originated separately from the aorta.

Endovascular multiple micro-coil embolization of the CHA, splenic artery (SA) and the aneurysm was done with variable sizes of IDC coils (Boston Scientific Co., Marlborough, MA, USA) and MicroNester coils (Cook Inc., Bloomington, IN, USA), followed by a stent-graft (Viabahn, 9x50 mm; W.L. Gore & Associates Inc., Flagstaff, AZ, USA) deployment in the SMA covering the orifice to the aneurysm. The postoperative course was uneventful with no signs of hepatic or splenic infarct. A postoperative coronal image of CT angiography resembles a gentle giraffe (Fig. 1B, C). Follow-up CT angiography after 1 year showed patent SMA stent-graft and well-occluded CMT aneurysm.

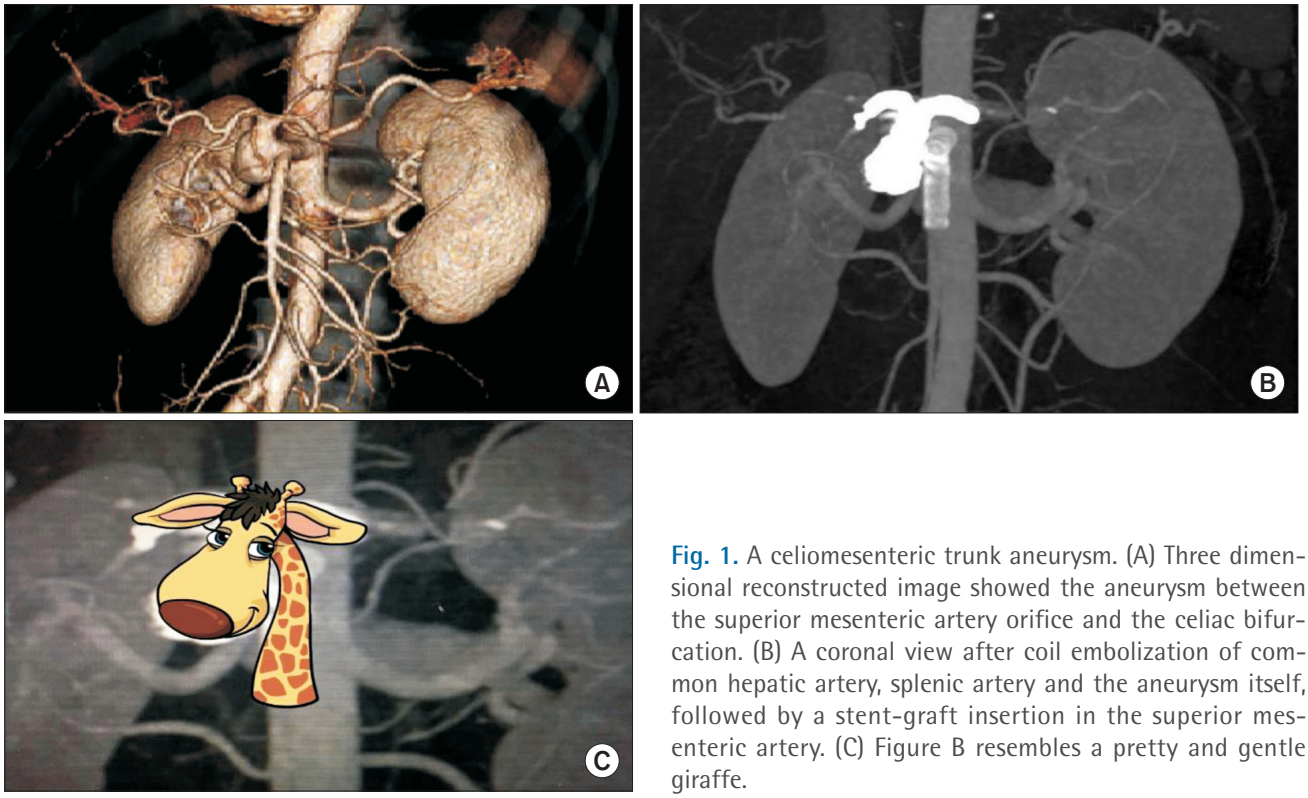

Fig. 1. A celiomesenteric trunk aneurysm. (A) Three dimensional reconstructed image showed the aneurysm between the superior mesenteric artery orifice and the celiac bifurcation. (B) A coronal view after coil embolization of common hepatic artery, splenic artery and the aneurysm itself, followed by a stent-graft insertion in the superior mesenteric artery. (C) Figure B resembles a pretty and gentle giraffe.

DISCUSSION

Visceral artery aneurysm is a rare disease, and the reported incidence is 0.1% to 0.2% of the population [1,2]. A CMT aneurysm is even rarer. A literature search revealed only 21 cases during the last 47 years [1-15], since Stanley et al. [3] reported the first case treated by open repair. All the reported cases were treated by open surgical repair. So this case, to our knowledge, is the first successful endovascular treatment of a CMT aneurysm ever reported.

Theoretically, a CMT is an embryologic error and may result in an increased risk for aneurysm formation due to the absence of a separate celiac trunk and an excessive blood influx into the origin of the CMT [2]. However, the exact relationship between the existence of a CMT and the aneurysm formation is not proven yet. During embryologic period, major visceral arteries develop from four vascular roots arising from the primitive dorsal abdominal aorta [2,4]. These four roots are united in utero by a ventral longitudinal anastomosis. During normal maturation, the first (gastric), second (hepatic), and the third (splenic) roots coalesce to form the main celiac axis, whereas the fourth root develops separately into the SMA. Alteration in the site of interruption of the ventral anastomosis will lead to the formation of a wide variety of vascular anomalies [16].

The exact natural history of this rare CMT aneurysm is unknown, but the recommendations for a visceral aneurysm can be applied to this rare disease. For the treatment of a CMT aneurysm, open surgical repair is the gold standard. The exact planning of the surgical procedures depends on the location, size, and the feature of the lesion. Preservation of the mesenteric and also hepatic circulation is the most important point. After resecting the aneurysm, direct anastomosis, reimplantation or bypass graft can be done according to the anatomy of each patient.

For selected cases, minimal invasive endovascular therapy can be applied. If feasible, a stent-graft excluding the CMT aneurysm is optimal, but usually the distance from the SMA orifice and/or from celiac bifurcation is not enough to prevent type Ia or type Ib endoleak from the landing zone after stent-graft deployment. Endovascular embolization of the aneurysm was often rejected due to the risk of ischemia to the liver and spleen, which would cause painful infarction.

However in this case, initial angiographic evaluation confirmed the good collateral circulations to the liver and spleen. Hepatic blood flow was maintained by collaterals via GDA, and splenic blood flow via short gastric arteries.

Because of the close proximity of the CMT aneurysm to the pancreas, there is a high risk of pancreatic fistula and bleeding from multiple pancreatic branches during open repair. Also there is a risk of diarrhea due to celiac plexus

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injury, pancreatitis, and injury to the surrounding viscera, including the portal venous system. Dissection of anomalous hepatic or SA from SMA may be difficult because they run into the pancreatic head neural plexus or the dorsal side of the pancreas.

Although the immediate result in this case is satisfactory, we do not know the long-term patency of the SMA stent-graft. Therefore long-term follow-up is mandatory for this kind of endovascular therapy.

A literature review of the CMT aneurysm written in English is summarized in Table 1. A total of 22 cases including our case has been reported. Interestingly 15 cases were from Far East Asia (Korea, Japan, and China), and 7 cases were from Western countries (USA, Italy, and France). Excluding a case series of Wang et al. [15] with insufficient data, the characteristics of the CMT aneurysm were analyzed (n=15). The morphology was mainly saccular in 12 (80%), or fusiform in 3 (20%). The location of the aneurysm was most common in the celiac trunk in 11 (73%), followed by CMT bifurcation in 3 and CMT orifice in 1. All operated cases were treated by aneurysmectomy and revascularization, except one case of endoaneurysmal Dacron patch repair [4]. Revascularization was done by direct anastomosis of celiac trunk to CMT in 7, bypass or interposition graft in 3, primary repair of aneurysm neck in 2, and reimplantation of celiac trunk into aorta in 1. Theoretically high flow in the conjoined CMT can be the cause of aneurysm and the separation of the celiac trunk and SMA may be prudent, but this procedure increases the complexity and risk of operation. Interestingly, in the literature, separation of the celiac trunk and SMA was done only in 1 case with reimplantation of celiac trunk to aorta [2]. No case of recurrent aneurysm has been reported whether the separation was done or not.

For treating this rare aneurysm, we should follow the basic principles for the treatment of visceral artery aneurysms. Furthermore, careful planning of surgical procedures according to the location, size, and the feature of the lesion, as well as collateral circulation are needed.

In conclusion, this report showed the world-first case of a successful endovascular treatment of a CMT aneurysm. Endovascular therapy can be done safely in selected cases of CMT aneurysm with sufficient collaterals to the liver and spleen.

Table 1. Reported cases of CMT aneurysm

First author, year	Country	Case	Age (y)	Sex	Shape	Size (mm)	Location	Revascularization
Stanley et al., 1970 [3]	USA	1	48	M	Fusiform	NS	CMT orifice	Bypass
Bailey et al., 1991 [4]	USA	1	46	F	Saccular	60	Celiac trunk	Endoaneurysmal Dacron patch repair
Detroux et al., 1998 [5]	France	1	51	M	Saccular	50	Bifurcation	Primary repair of aneurysm neck
Matsumoto et al., 1999 [6]	Japan	1	53	M	Fusiform	40	Bifurcation	Interposition graft, SA reimplantation
Kalra et al., 2003 [7]	USA	1	52	F	Saccular	20	Celiac trunk	Direct anastomosis
Matsuda et al., 2006 [8]	Japan	1	36	M	Saccular	NS	Celiac trunk	Direct anastomosis
Mammano et al., 2009 [9]	Italy	1	51	M	Saccular	33	Celiac trunk	Primary repair of aneurysm neck
Iida et al., 2010 [10]	Japan	1	46	M	Saccular	30	Celiac trunk	Direct anastomosis
Obara et al., 2009 [11]	Japan	1	73	M	Fusiform	25	CMT	Bypass
Wang et al., 2010 [12]	China	1	53	M	Saccular	38	Celiac trunk	Primary repair & reimplantation of SMA to celiac trunk
Guntani et al., 2011 [13]	Japan	1	82	M	Saccular	25	Celiac trunk	Direct anastomosis
Higashiyama et al., 2011 [14]	Japan	1	54	M	Saccular	40	Celiac trunk	Direct anastomosis
Wang et al., 2014 [15]	China	7	-	-	NS	NS	NS	NS
Lipari et al., 2015 [2]	Italy	1	52	M	Saccular	30	Celiac trunk	Reimplantation of celiac trunk to aorta
VonDerHaar et al., 2015 [1]	USA	1	45	M	Saccular	30	Celiac trunk	Direct anastomosis
Current Study	Korea	1	45	M	Saccular	21	Celiac trunk	Endovascular coil embolization, SMA stent-graft

CMT, celiomesenteric trunk; –, not available; M, male; F, female; NS, not stated; SA, splenic artery; SMA, superior mesenteric artery.

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