Celiac artery embolism due to thrombophilia – a case report

George Trellopoulos MD¹, Maria I Pikilidou MD MSc², Evaggelia Tsiga MD MSc²

G Trellopoulos, MI Pikilidou, E Tsiga. Celiac artery embolism due to thrombophilia – a case report. *Int J Angiol* 2009;18(2):96-98.

A case of celiac artery embolism in a patient with factor V Leiden thrombophilia is reported. The embolism was likely due to an undetected cardiac thrombus, causing an abdominal aortic embolism. The

patient underwent emergency surgery for the abdominal embolism. The celiac artery embolism was treated nonsurgically due to the presence of collateral circulation through the gastroduodenal artery from the superior mesenteric artery. The patient fully recovered and was discharged from the hospital one month after his first referral.

Key Words: *Celiac artery embolism; Factor V Leiden; Thrombophilia*

Celiac artery occlusion is a potentially critical surgical condition that can cause intestinal malperfusion and ischemia to multiple organs, such as the liver, stomach, pancreas and spleen. It can be caused by compression from the median arcuate ligament (1-5); intravascularly from atherosclerosis (2,6) and embolism (2,7); and extrinsically from lymphadenopathy (2), direct tumour extension (2,8), intimal hyperplasia (7) and inferior pancreaticoduodenal artery aneurysm compression (9,10), as well as injury from previous catheter manipulation or surgical trauma (8). Moreover, occlusion can also occur during pancreaticoduodenectomy (2,11) and endovascular treatment of type 2 dissection. There is a report in the literature of compression from superior mesenteric artery (SMA) dissection (12). Celiac stenosis or occlusion can also be a congenital anatomical abnormality (1).

We report a case of celiac artery occlusion caused by massive arterial embolism due to thrombophilia, which was triggered by cessation of acenocoumarol and treated nonsurgically.

CASE PRESENTATION

A 48-year-old man was referred to Papanikolaou General Hospital (Thessaloniki, Greece) due to pleural pain, which started three days before referral. He also reported an episode of hemoptysis two days earlier. The patient had a medical history of factor V Leiden thrombophilia (heterozygous), which had produced a myocardial infarction 15 years previously and an ischemic stroke one year previously. Acenocoumarol was used for the treatment of thrombophilia. His chest x-ray showed perfusion of the left upper lobe with possible central necrosis close to the left portal. A laboratory test was positive for D-dimers (0.766 µg/mL, normal range 0 µg/mL to 0.500 µg/mL). A pulmonary embolism was suspected but was excluded by a computed tomography (CT) angiogram. Bronchoscopy was also performed for diagnostic purposes. For prevention of hemorrhage, the acenocoumarol was discontinued two days before the examination and was replaced with low molecular weight heparin. After the procedure, the patient showed signs of

abdominal aortic embolism (low back pain, discolouration of the lower extremities, paralysis and absence of pulse). The pre-operative angiogram showed not only an embolus in the aorta but also occlusion of the celiac artery, branches of the SMA and distal branches of the renal arteries (Figures 1 and 2). He underwent emergency embolectomy of the aorta by the Fogarty method from the femoral arteries, according to standard procedure. A nonsurgical approach to the celiac artery embolism was chosen due to the absence of abdominal complaints, the severe general condition of the patient, the absence of parenchymal ischemia and the presence of collateral circulation through the gastroduodenal artery from the SMA, as shown on the angiogram. The patient fully recovered from surgery and was discharged after one month, with normal liver and renal function. The patient was closely monitored for complications. A CT angiogram three months later verified the collateral circulation through the gastroduodenal artery (Figure 3).

DISCUSSION

Our patient suffered from a known abnormality of hemostasis associated with pathological thrombus formation caused by a mutation in the gene coding factor V, also known as factor V Leiden (13), which is present in 4% to 6% of the general population (14). In individuals heterozygous for factor V Leiden, the activated partial thromboplastin time is moderately decreased, whereas in homozygous individuals there is little change at all (15). Though factor V Leiden is associated with high risk for primary and recurrent venous thromboembolism, by itself it does not seem to increase the risk for arterial embolism (16).

Our patient was undergoing treatment with acenocoumarol for the thrombophilia, but this had been discontinued. Therefore, the most probable cause of the massive arterial embolism that occurred seems to have been the formation of a cardiac thrombus that remained undetected.

Population studies have shown a prevalence between 1% and 12.5% for celiac artery occlusion in the general

¹Department of Thoracic Surgery; ²Second Department of Internal Medicine, Papanikolaou General Hospital, Thessaloniki, Greece
Correspondence: Dr Maria I Pikilidou, 2nd Department of Internal Medicine, Papanikolaou General Regional Hospital, Exohi 57010, Thessaloniki, Greece. Telephone 30-231-021-7760, fax 30-231-021-7760, e-mail m_pikilidou@yahoo.com

Figure 1) Computed tomography angiogram before (A) and after (B) revascularization of the aorta. Arrows indicate the embolus in the aorta

Figure 2) Computed tomography angiogram before (left panel) and after (right panel) revascularization of the aorta. Upper arrow: Tripod of Haller; Lower arrow: Thrombus in the aorta

population, and a prevalence of 1% to 24% for celiac axis stenosis (2,6-10,12).

Most of these occlusions, while they may be significant radiographically, produce no abdominal complaints, due to the development of collateral circulation from the gastroduodenal artery, the pancreaticoduodenal arcades and the dorsal pancreatic artery from the SMA (2). This generally occurs when the occlusion is caused gradually.

On the other hand, in cases of acute embolism, visceral ischemia can cause symptoms such as abdominal pain, associated nausea, vomiting and frequent bowel movements (16). Abdominal pain is present in most patients with acute celiac axis occlusion, although it is absent in some cases.

Angiography is the gold standard in the diagnosis of celiac axis occlusion (17). By means of angiography, collateral flow to the common hepatic artery can be also seen as retrograde flow from the SMA via a dilated gastroduodenal artery or dorsal pancreatic artery (2,3,8,17).

Figure 3) Computed tomography angiogram three months after the referral. A Celiac artery occlusion; B Collateral circulation through the gastroduodenal artery

A CT angiogram can also diagnose embolisms but with lower sensitivity. A study of 26 patients showed that biphasic CT with mesenteric CT angiography was 100% specific, but only 73% sensitive in the diagnosis of SMA occlusion, celiac and inferior mesenteric artery occlusion, and arterial embolism (4).

Perioperatively, the diagnosis of celiac artery occlusion is possible using the decrease or absence of pulsations in the hepatic artery, intraoperative ultrasonography, and observing persistent discolouring of the hepatic parenchyma. The diagnosis is critical, especially for pancreatic surgeons, to prevent severe complications during pancreatectomy and endovascular repair of descending thoracic aneurysms with the use of a thoracic stent (11).

The treatment options for celiac artery occlusion depend on the severity of the occlusion, the development of collateral circulation and the patient's clinical condition.

Endovascular treatment of celiac and mesenteric stenosis has a low incidence of complications, lower morbidity and a high technical success rate (18).

Operative therapy is used as a treatment for celiac axis occlusion due to median arcuate ligament syndrome. Takach et al (19) reported a study of seven patients who underwent tailored operative intervention (decompression alone or with revascularization) for celiac artery compression syndrome, and achieved immediate and long-term relief from the symptoms in all patients. In another study (2), 14 patients who underwent pancreaticoduodenectomy and had celiac axis occlusion due to median arcuate ligament syndrome were treated successfully with a combination of median arcuate ligament release, vascular reconstruction and/or endovascular celiac stenting, and no surgical mortalities or postoperative complications occurred in six patients (46%).

On the other hand, there are some reports of celiac artery occlusions that were treated nonsurgically. In a study of three cases of celiac artery embolism synchronous with SMA embolism, only one required revascularization due to discolouration of the hepatic parenchyma (7). Furthermore, one case of celiac artery occlusion caused by acute type B aortic dissection

that was treated nonsurgically was subjected to resection of the small intestine three weeks later because of ulcer perforation due to ischemia (5). Moreover, celiac artery coverage during endovascular thoracic aortic aneurysm repair in seven patients did not show any mesenteric ischemic complications. In six of these patients, collateral circulation through the gastroduodenal artery was demonstrated preoperatively (20). Also, in a survey of 15 patients with celiac axis occlusion who underwent pancreaticoduodenectomy, trial clamping of the gastroduodenal artery was performed in 11 patients, and provoked obvious ischemia in only four (11).

Finally, in a study of 11 patients with celiac axis stenosis (21) who underwent resection of the pancreatic head for chronic pancreatitis, division of the median arcuate ligament was performed to re-establish hepatic artery blood flow in only two cases.

We conclude that a thorough perioperative assessment of the viscera is essential in deciding whether to use a surgical approach to celiac artery occlusion. Imaging is very helpful in assessing collateral circulation and should be used as a guide. The present case serves as a reminder to vascular surgeons to avoid unnecessary revascularization of the celiac artery when there is adequate collateral circulation. Also, screening for cardiac thrombi before any intervention should always be considered to avoid embolism.

REFERENCES

1. Bech F, Loesberg A, Rosenblum J, Glagov S, Gewertz BL. Median arcuate ligament compression syndrome in monozygotic twins. *J Vasc Surg* 1994;19:934-8.
2. Farma JM, Hoffman JP. Nonneoplastic celiac axis occlusion in patients undergoing pancreaticoduodenectomy. *Am J Surg* 2007;193:341-4.
3. Horton KM, Talamini MA, Fishman EK. Median arcuate ligament syndrome: Evaluation with CT angiography. *Radiographics* 2005;25:1177-82.
4. Kirkpatrick ID, Kroeker MA, Greenberg HM. Biphasic CT with mesenteric CT angiography in the evaluation of acute mesenteric ischemia: Initial experience. *Radiology* 2003;229:91-8.
5. Takahashi K, Takahashi S. [Treatment of acute type B aortic dissection with organ ischemia.] *Kyobu Geka* 2007;60:303-7.
6. Cornell SH. Severe stenosis of the celiac artery. Analysis of patients with and without symptoms. *Radiology* 1971;99:311-6.
7. Park CM, Chung JW, Kim HB, Shin SJ, Park JH. Celiac axis stenosis: Incidence and etiologies in asymptomatic individuals. *Korean J Radiol* 2001;2:8-13.
8. Kwon JW, Chung JW, Song SY, et al. Transcatheter arterial chemoembolization for hepatocellular carcinomas in patients with celiac axis occlusion. *J Vasc Interv Radiol* 2002;13:689-94.
9. Ikeda O, Tamura Y, Nakasone Y, Kawanaka K, Yamashita Y. Coil embolization of pancreaticoduodenal artery aneurysms associated with celiac artery stenosis: Report of three cases. *Cardiovasc Intervent Radiol* 2007;30:504-7.
10. Peterson BG, Resnick SA, Eskandari MK. Coil embolization of an inferior pancreaticoduodenal artery aneurysm associated with celiac artery occlusion. *Cardiovasc Surg* 2003;11:515-9.
11. Berney T, Pretre R, Chassot G, Morel P. The role of revascularization in celiac occlusion and pancreaticoduodenectomy. *Am J Surg* 1998;176:352-6.
12. Lindner HH, Kemprud E. A clinicoanatomical study of the arcuate ligament of the diaphragm. *Arch Surg* 1971;103:600-5.
13. Greengard JS, Eichinger S, Griffin JH, Bauer KA. Brief report: Variability of thrombosis among homozygous siblings with resistance to activated protein C due to an Arg->Gln mutation in the gene for factor V. *N Engl J Med* 1994;331:1559-62.
14. Ridker PM, Miletich JP, Stampfer MJ, Goldhaber SZ, Lindpaintner K, Hennekens CH. Factor V Leiden and risks of recurrent idiopathic venous thromboembolism. *Circulation* 1995;92:2800-2.
15. Rosendaal FR, Koster T, Vandenbroucke JP, Reitsma PH. High risk of thrombosis in patients homozygous for factor V Leiden (activated protein C resistance). *Blood* 1995;85:1504-8.
16. Price DT, Ridker PM. Factor V Leiden mutation and the risks for thromboembolic disease: A clinical perspective. *Ann Intern Med* 1997;127:895-903.
17. Patten RM, Coldwell DM, Ben-Menachem Y. Ligamentous compression of the celiac axis: CT findings in five patients. *AJR Am J Roentgenol* 1991;156:1101-3.
18. Mohammed A, Teo NB, Pickford IR, Moss JG. Percutaneous transluminal angioplasty and stenting of coeliac artery stenosis in the treatment of mesenteric angina: A case report and review of therapeutic options. *J R Coll Surg Edinb* 2000;45:403-7.
19. Takach TJ, Livesay JJ, Reul GJ Jr, Cooley DA. Celiac compression syndrome: Tailored therapy based on intraoperative findings. *J Am Coll Surg* 1996;183:606-10.
20. Vaddineni S, Taylor S, Patterson M, et al. Outcome after celiac artery coverage during endovascular thoracic aortic aneurysm repair: Preliminary results. Sixteenth Annual Winter Meeting of the Peripheral Vascular Surgery Society. Park City, January 26 to 29, 2006.
21. Pfeiffenberger J, Adam U, Drognitz O, et al. Celiac axis stenosis in pancreatic head resection for chronic pancreatitis. *Langenbecks Arch Surg* 2002;387:210-5.