Spontaneous dissection of the celiac trunk : a rare cause of abdominal pain – case report and review of the literature

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Abstract

Spontaneous dissection of the celiac trunk is uncommon and rarely considered in patients presenting with acute onset of epigastric pain. We report the case of a 48-year old male, diagnosed with a spontaneous dissection of the celiac trunk extending towards the common hepatic artery. He was treated conservatively and remained asymptomatic after two years of follow-up. Conservative treatment seems justified in the absence of bowel ischemia or signs of hemorrhage. Initial Computed Tomography angiography revealed the presence of a dissection with a pseudoaneurysm that remained stable and regressed towards a normal Computed Tomography angiography after 7 months of follow-up. Radiologic follow-up is warranted as progression of the dissection and/or total occlusion with or without symptoms can occur. The risk factors, the natural course and optimal treatment remain unclear due to the rarity of the disorder. Our patient had no predisposing cardiovascular risk factors. Nevertheless, we observed a hypertrophic ligamentum arcuatum on Computed Tomography, possibly facilitating the evolution towards a dissection. Next to the case report, we provide a review of the available literature. (Acta gastroenterol. belg., **2013**, 76, **335-339**).

Key words : dissection, celiac trunk, truncus celiacus, hepatic artery, abdominal pain.

Introduction

Spontaneous dissection of the celiac trunk is a rare finding and an uncommon cause of abdominal pain. Only case reports and a few case series, all retrospective analyses, have been published (1-5). Therefore the risk factors and natural course remains unclear. Therapeutic strategies and follow-up procedures are based on limited observations. Here we describe a case, presenting with acute onset of abdominal pain resulting from a spontaneous dissection of the celiac trunk. Next, we provide a review of the existing literature.

Case report

A 48-year old male presented at the emergency department because of a sudden onset of epigastric pain. The pain started immediately after a high caloric lunch, had a stabbing nature and was irradiating towards his back. Upon admission, his initial pain had already spontaneously decreased (visual analog scale score from 8/10 to 5/10). He reported no trauma, nausea or an urge to move. He had a history of a sliding hiatal hernia with reflux oesophagitis for several years for which he was taking proton pump inhibitors, but indicated this was a different kind of pain. He stopped smoking 4 years before and had no other cardiovascular risk factors. His family history was negative.

At presentation his blood pressure was 130/70 mmHg and abdominal palpation revealed a mild tenderness over the epigastrium. Physical examination was otherwise normal. After administration of paracetamol (1 g) and butylhyocin (20 mg) he became asymptomatic. Initial blood tests including C-reactive protein, cardiac enzymes, complete blood count, renal function tests, liver enzymes, amylase, lipase, d-dimers and urine analysis were normal. Additional auto-immune serology was negative. Electrocardiogram (ECG) and ultrasound examination of the abdomen demonstrated no abnormalities. The day after admission, an esophagogastroscopy did not reveal any evidence for active reflux disease. A contrast enhanced Computed Tomography (CT) of the abdomen however demonstrated a diffuse infiltration along the celiac trunk and hepatic artery (Fig. 1A), identified using magnetic resonance imaging and CT angiography (Fig. 1B) as an isolated dissection. No perfusion defects were observed and the left gastric and splenic artery were normal. A hypertrophic ligamentum arcuatum was visualized, suggesting a possible relationship with the dissection. The patient was managed conservatively, received no antiplatelet or anticoagulant therapy and had no recurrence of symptoms. A follow-up CT Angiography performed after 4 and 12 weeks demonstrated a persistent dissection of the celiac trunk and hepatic artery with the presence of a stable pseudoaneurysm of 11 mm (Fig. 1B, arrow). CT angiography performed after 7 and 18 months demonstrated a normal celiac trunk without evidence of dissection or pseudoaneurysm formation (Fig. 2). These findings were confirmed after 24 months when the patient was readmitted at the emergency department because of upper abdominal pain. After a local infiltration with xylocaine at the xyphoid processus, his symptoms resolved confirming the musculoskeletal origin of his pain. He remains in good health two years after diagnosis.

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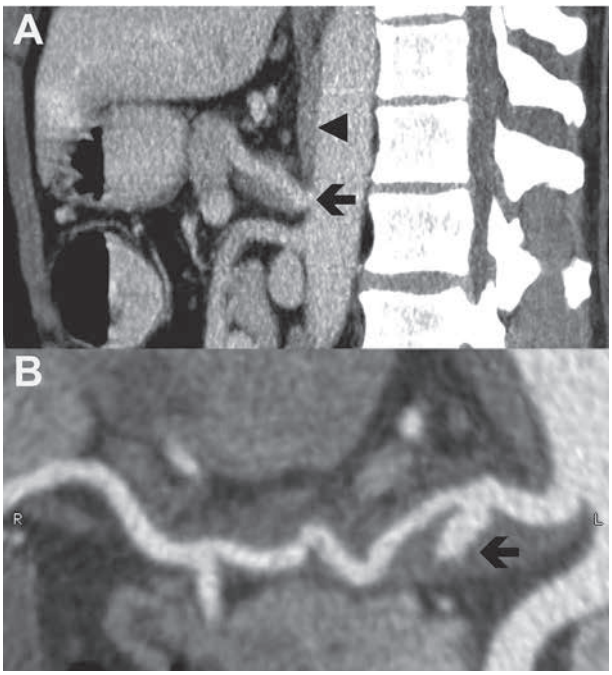

Fig. 1. — (A) Contrast enhanced CT of the abdomen, sagital section demonstrating the dissection of the celiac trunk (arrow) and hypertrophic ligamentum arcuatum (arrowhead). (B) CT angiography demonstrating the dissection of the celiac trunk with the presence of a pseudoaneurysm of 11 mm, just before the split towards the common hepatic and splenic artery (arrow).

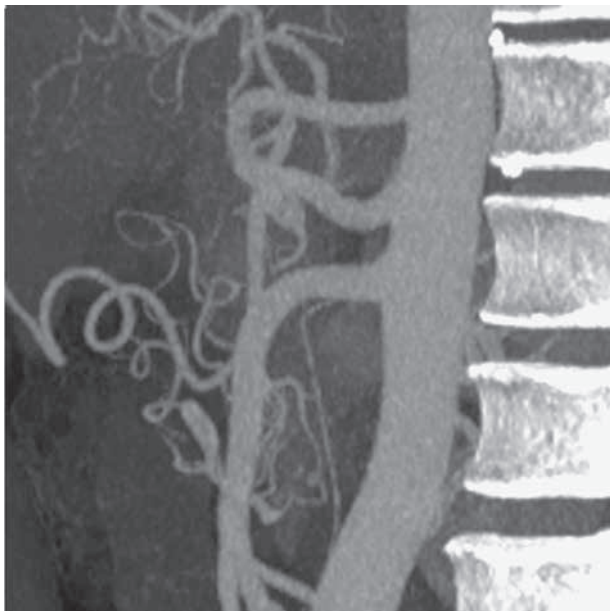

Fig. 2. — CT angiography image at 7 months of follow-up demonstrating the absence of signs of dissection or pseudoaneurism.

Discussion

Isolated dissection of the celiac trunk is a rare finding and a pitfall in the diagnosis of abdominal pain. Review of the available English literature demonstrates a total of 78 cases, including ours, described so far (listed in Table 1). Median age was 54 years (range 28 to 89) and 77% were male. The majority of cases presented with epigastric or upper abdominal pain. In 15 cases (19%) the dissection was found incidentally after radiologic evaluation performed for other reasons. Both an acute onset of pain and chronic abdominal pain are reported. Also obstructive jaundice due to compression by an aneurismal dilatation of the dissection extending to the hepatic artery (6), and intestinal angina (7) have been described.

Reported risk factors are those for peripheral artery dissections and include atherosclerotic disease, arterial hypertension, fibromuscular dysplasia, cystic medial necrosis, periarterial inflammation in association with cholecystitis or pancreatitis, trauma, pregnancy, and connective tissue diseases (8). Recently a dissection following a valsalva maneuver in a weight lifter was reported (9). However, most cases presents without any identifiable risk factor (5,10). Isolated dissection of the superior mesenteric artery parallels this observation. In a series of 22 patients with an isolated dissection of the superior mesenteric artery described by Yasuhara *et al.,* the estimated etiologic factors were atherosclerosis in 4 (18%), medial necrosis or degeneration in 3 (14%), suspected fibromuscular dysplasia in 1 (5%), and unknown in 14 (63%) (11). Our patient had no predisposing risk factors besides a history of smoking. Whether the presence of a hypertrophic ligamentum arcuatum in our patient caused or facilitated the evolution towards a dissection remains uncertain. This is however the first report, to our knowledge, reporting the presence of both. A hypertrophic ligamentum arcuatum can lead to a celiac artery compression syndrome. This is associated with a variety of symptoms including postprandial pain and weight loss (12) but its relevance remains controversial (reviewed in (13)). Our patient had complaints of epigastric pain, attributed to reflux oesophagitis, long before his presentation with a dissection of the celiac trunk. A relationship with these complaints and a possible celiac artery compression syndrome is unknown. We cannot rule out progression from an earlier lesion as our patients had undergone no abdominal imaging before.

Isolated dissection of the celiac trunk was historically associated with a poor prognosis (13,14). Since 1989 however, no fatalities have been reported probably not reflecting a changing pattern but rather improved diagnostic screening techniques (15) and/or a hesitancy to report adverse outcomes (4). The natural history remains unpredictable, but spontaneous resolution, definitive occlusion, aneurysm formation, or rupture of a visceral artery can occur. Acute signs of bleeding or liver ischemia are poor prognostic factors (2,16). Some case reports indicate reoccurrence of symptoms or progression of the dissection within the first months after the initial presentation, warranting radiographic follow-up (2,4,10,17). However the exact duration of this follow-up remains uncertain. Of note, the psychological burden associated

Table 1

Author	Year	Number of patients (Male/ Female)	Mean age (range)	Symptoms (number of patients)	Treatment	Follow-up
Pinkerton *et al*. (20)	1976	1 (1/0)	52	Abdominal pain	Resection and graft	18 months ; good outcome
Larson *et al*. (14)	1959-1987	10 (3/7)	57 (46-70)	Asymptomatic (2), abdominal pain (8)	None	postmortem diagnosis (10) : acute rupture (4), unrelated death cause (6)
Bret *et al*. (6)	1987	1 (1/0)	61	Upper abdominal pain, jaundice	Ligation of hepatic artery	Died a few days later (missed Debakey type III aortic dissection)
Roh *et al*. (19)	1989	1 (0/1)	65	Acute epigastric pain	None	Postmorten diagnosis, acute rupture
Bartoli *et al*. (21)	1990	1 (0/1)	32	Epigastric pain ; occlusion of hepatic and splenic arteries	Aortohepatic and aortosplenic bypass	Not reported (Angiographic control patent bypasses)
Müller *et al*. (22)	1995	2 (1/1)	50 (36-61)	Abdominal pain and shock due to ruptured hepatic artery, lower back pain	Conservative, antihypertensives (1) ; Saphenous vein bypass (1)	3-8 years ; good outcome
Takeda *et al*. (18)	1995	1 (1/0)	61	Acute abdominal pain	Coil embolisation	Not reported ; transient cholecystitis
Chaillou *et al*. (7)	1997	1 (0/1)	64	Intestinal angina	Prosthetic bypass to SMA and CA	6 months ; good outcome
Hashimoto *et al*. (23)	1998	1 (0/1)	28	Acute epigastric pain	Conservative	6 months ; good outcome
Matsuo *et al*. (24)	2000	1 (1/0)	58	Upper abdominal pain	Conservative	Not reported
Glehen *et al*. (2)	2001	5 (3/2)	54 (46-72)	Asymptomatic (1), acute abdominal pain (3), chronic epigastric pain and weight loss (1)	Conservative (2) ; Resection and bypass, interposition graft or reanastomosis (3),	14 (6-24) months ; good outcome
Fenoglio *et al*. (17)	2004	2 (2/0)	48 (41-54)	Acute abdominal pain	Conservative (1) ; resection and reanastomosis (1)	6 months ; good outcome
Batsis *et al*. (16)	2005	1 (0/1)	67	Postprandial abdominal pain	Prosthetic bypass	Not reported
McGuinness *et al*. (25)	2006	1 (1/0)	57	Acute epigastric pain	Conservative	3 months ; CTA stable
D'Ambrosio *et al*. (1)	2007	6 (6/0)	59 (45-89)	Asymptomatic (1), abdominal pain (5)	Conservative (6)	1 year ; good outcome
Woolard *et al*. (26)	2007	1 (1/0)	53	Lower abdominal pain and nausea	Conservative ; anticoagulant therapy	9 months ; CTA stable dissection
Poylin *et al*. (27)	2008	1 (1/0)	60	Acute abdominal pain	Conservative ; anticoagulant therapy	6 months ; good outcome
Takayama *et al*. (3)	2008	6 (4/2)	53 (44-66)	Asymptomatic (4), abdominal pain (2)	Conservative, no antiplatelet/other (4), anticoagulant therapy (1) ; Surgical repair (1)	14 (2-30) months ; good outcome
Nordanstig *et al*. (28)	2008	1 (1/0)	56	Acute abdominal pain ; rupture of splenic artery	Resection and splenectomy	6 months ; CTA stable
Basile *et al*. (29)	2009	1 (0/1)	67	Asymptomatic (hepatocellular carcinoma)	Stenting to allow future chemoembolisation	Not reported
Takach *et al*. (4)	2009	6	55 (42-75)	Acute abdomoinal pain	Conservative ; endovascular or operative intervention	13 (2-36) months ; good outcome
Mousa *et al*. (30)	2009	1 (1/0)	57	Epigastric abdominal pain	Conservative, antiplatelet, anticoagulant therapy	18 months ; good outcome

Wang *et al*. (31)	2010	1 (1/0)	44	Acute upper abdominal pain	Conservative ; antihypertensive therapy	6 months ; good outcome
Tokue *et al*. (5)	2009	10	61	Asymptomatic (6), abdominal pain (4)	Conservative	
Ozaki *et al*. (32)	2010	1 (1/0)	50	Abdominal pain	Conservative, aspirine, antihypertensive therapy	10 months ; good outcome
Zeina *et al*. (33)	2010	1 (1/0)	49	Acute abdominal pain	Conservative	Not reported
Oh *et al*. (10)	2010	8 (8/0)	46 (39-57)	Acute abdominal pain	Conservative (aspirin and/or anticoagulant therapy until follow-up CT scan demonstrated regression or stabilisation) (7) ; endovascular stent (1)	16 months ; good outcome
Riles *et al*. (9)	2011	1 (1/0)	45	Abdominal pain after weight lifting	conservative ; anticoagulant therapy	6 months ; CTA stable
Batt *et al*. (15)	2011	1 (1/0)	43	Acute upper abdominal pain	Percutaneous embolisation	9 months ; CTA stable
Ozturk *et al*. (34)	2011	1 (1/0)	40	Stabbing epigastric pain	Conservative, antihypertensive therapy	10 months ; good outcome
Zhang et al. (35)	2012	1 (1/0)	44	Abdominal pain, vomiting	Endovascular stenting	2 months ; good outcome

Abbreviations : SMA : superior mesenteric artery ; CA : celiac artery ; CTA : CT angiography.

with the unpredictable outcome should not be underestimated. In our case this led to two readmissions triggered by mild abdominal complaints.

Initial cases were mostly diagnosed post-mortem (14). Nowadays, the widespread use of contrast enhanced CT scanning has definitely enabled earlier and more accurate diagnosis. In our case also MRI proved to be valuable in the diagnostic approach.

The unknown natural course makes the therapeutic decision process highly challenging (3). Treatment may involve surveillance or a surgical and/or endovascular repair (2,11).

Surgical treatment has been recommended for patients presenting with (a) signs of rupture, (b) ischemia, (c) persistent or recurrence of symptoms, (d) the presence of a fusiform or sacculated aneurysm with increasing diameter, or (e) thrombosis of the true lumen of the dissection site (2). The involvement of the hepatic artery as an indication for surgery (18), because of the suggested high mortality (2,19), remains somewhat controversial given the increasing number of conservatively treated cases with favorable outcome (1,3-5,10). As in our case, conservative medical treatment can be proposed for patients with limited dissection in whom serial examinations have demonstrated no evidence of rupture or expansion (1,3,5). Cardiovascular risk factor modification, limiting the propagation of the dissection and reducing the risk for rupture, is warranted. The role of antiplatelet or anticoagulant therapy to prevent thromboembolic complications is a matter of ongoing debate. The use of anticoagulant or antiplatelet agents for 3 to 6 months with a target international normalized ratio of 2.0 to 3.0, has been suggested (17). Recently, the use of anticoagulant therapy until a follow-up CT demonstrates regression or an unchanged diameter of the false lumen, resulted in good clinical outcome even though a subset of patients presented with signs of ischemia (10). Others however suggested equal outcome even in the absence of anticoagulant or antiplatelet therapy (5).

In conclusion, our case report of an isolated dissection of the celiac trunk represents a rare cause of abdominal pain. The absence of guidelines warrants a multidisciplinary and individualized approach.

References

1. D'AMBROSIO N., FRIEDMAN B., SIEGEL D., KATZ D., NEWATIA A., HINES J. Spontaneous isolated dissection of the celiac artery : CT findings in adults. *AJR. Am. J. Roentgenol*., 2007, **188** : W506-W511.
2. GLEHEN O., FEUGIER P., ALEKSIC Y., DELANNOY P., CHEVALIER J.M. Spontaneous dissection of the celiac artery. *Ann. Vasc. Surg*., 2001, **15** : 687-692.
3. TAKAYAMA T., MIYATA T., SHIRAKAWA M., NAGAWA H. Isolated spontaneous dissection of the splanchnic arteries. *J. Vasc. Surg*., 2008, **48** : 329-333.
4. TAKACH T.J., MADJAROV J.M., HOLLEMAN J.H., ROBICSEK F., ROUSH TS. Spontaneous splanchnic dissection : application and timing of therapeutic options. *J. Vasc. Surg*., 2009, **50** : 557-563.
5. TOKUE H., TSUSHIMA Y., ENDO K. Imaging findings and management of isolated dissection of the visceral arteries. *Jpn. J. Radiol*., 2009, **27** : 430-437.
6. BRET P.M., PARTENSKY C., BRETAGNOLLE M., PALIARD P., BURKE M. Obstructive jaundice by a dissecting aneurysm of celiac axis and hepatic artery. *Dig. Dis. Sci*., 1987, **32** : 1431-1434.
7. CHAILLOU P., MOUSSU P., NOEL S.F., SAGAN C., PISTORIUS M.A., LANGLARD J.M. *et al*. Spontaneous dissection of the celiac artery. *Ann. Vasc. Surg*. 1997, **11** : 413-415.

8. LOK S.Y., CHALVARDJIAN A., COMMON A.A. Primary renal artery dissection. *Can. Assoc. Radiol. J.*, 1995, **46** : 54-56.
9. RILES T.S., LIN J.C. Celiac artery dissection from heavy weight lifting. *J. Vasc. Surg.*, 2011, **53** : 1714-1715.
10. OH S., CHO Y.P., KIM J.H., SHIN S., KWON T.W., KO G.Y. Symptomatic spontaneous celiac artery dissection treated by conservative management : serial imaging findings. *Abdom. Imaging*, 2010, **36** : 79-82.
11. YASUHARA H., SHIGEMATSU H., MUTO T. Self-limited spontaneous dissection of the main trunk of the superior mesenteric artery. *J. Vasc. Surg.*, 1998, **27** : 776-779.
12. SCHOLBACH T. Celiac artery compression syndrome in children, adolescents, and young adults : clinical and color duplex sonographic features in a series of 59 cases. *J. Ultrasound Med.*, 2006, **25** : 299-305.
13. LOUKAS M., PINYARD J., VAID S., KINSELLA C., TARIQ A., TUBBS R.S. Clinical anatomy of celiac artery compression syndrome : a review. *Clin. Anat.*, 2007, **20** : 612-617.
14. LARSON C.J., GEIER G.R. JR., EDWARDS W.D. Fatal acute dissection of the right hepatic artery after appendectomy. *Arch. Pathol. Lab. Med.*, 1987, **111** : 300-302.
15. BATT M., BAQUE J. Successful percutaneous embolization of a symptomatic celiac artery dissection with aneurysmal dilation with detachable vascular plugs. *J. Vasc. Surg.*, 2011, **54** : 1812-1815.
16. BATSIS J.A., ARORA A.S. Celiac artery dissection : an uncommon cause of abdominal pain and weight loss. *Clin. Gastroenterol. Hepatol.*, 2005, **3** : A30.
17. FENOGLIO L., ALLIONE A., SCALABRINO E., ALBERTO G., BENEDETTI V., POMERO F. *et al*. Spontaneous dissection of the celiac artery : a pitfall in the diagnosis of acute abdominal pain. Presentation of two cases. *Dig. Dis. Sci.*, 2004, **49** : 1223-1227.
18. TAKEDA H., MATSUNAGA N., SAKAMOTO I., OBATA S., NAKAMURA S., HAYASHI K. Spontaneous dissection of the celiac and hepatic arteries treated by transcatheter embolization. *AJR Am. J. Roentgenol.*, 1995, **165** : 1288-1289.
19. ROH L.S., LE SHER A. Dissecting aneurysm of the hepatic artery. *Am. J. Forensic. Med. Pathol.*, 1989, **10** : 67-70.
20. PINKERTON J.A. JR., WOOD W.G., FOWLER D. Fibrodysplasia with dissecting aneurysm of the hepatic artery. *Surgery*, 1976, **79** : 721-723.
21. BARTOLI J.M., MOULIN G., DI STEFANO D., RUDONDY P., GEROLAMI A., KASBARIAN M. Isolated dissection of the celiac trunk and its branches. X-ray computed tomography and angiography findings. A case report. *Ann. Radiol. (Paris)*, 1990, **33** : 264-266.
22. MULLER M.F., KIM D. Spontaneous dissection of the hepatic artery. *Abdom. Imaging*, 1995, **20** : 462-465.
23. HASHIMOTO M., ITO I., SATO K. Case report : Dissecting aneurysm of the hepatic artery. *Clin. Radiol.*, 1998, **53** : 913-914.
24. MATSUO R., OHTA Y., OHYA Y., KITAZONO T., IRIE H., SHIKATA T. *et al*. Isolated dissection of the celiac artery – a case report. *Angiology*, 2000, **51** : 603-607.
25. MC GUINNESS B., KENNEDY C., HOLDEN A. Spontaneous coeliac artery dissection. *Australas Radiol.*, 2006, **50** : 400-401.
26. WOOLARD J.D., AMMAR A.D. Spontaneous dissection of the celiac artery : a case report. *J. Vasc. Surg.*, 2007, **45** : 1256-1258.
27. POYLIN V., HILE C., CAMPBELL D. Medical management of spontaneous celiac artery dissection : case report and literature review. *Vasc. Endovascular Surg.*, 2008, **42** : 62-64.
28. NORDANSTIG J., GERDES H., KOCYS E. Spontaneous isolated dissection of the celiac trunk with rupture of the proximal splenic artery : a case report. *Eur. J. Vasc. Endovasc. Surg.*, 2009, **37** : 194-197.
29. BASILE A., TSETIS D., MONTINERI A., COPPOLINO F., PATTI M.T., FATUZZO F. Self-expanding stent placement as a bridge for safe hepatic chemoembolization in a patient with isolated spontaneous dissection of the celiac artery. *J. Vasc. Interv. Radiol.*, 2009, **20** : 425-426.
30. MOUSA A.Y., COYLE B.W., AFFUSO J., HASER P.B., VOGEL T.R., GRAHAM A.M. Nonoperative management of isolated celiac and superior mesenteric artery dissection : case report and review of the literature. *Vascular*, 2009, **17** : 359-364.
31. WANG J.L., HSIEH M.J., LEE C.H., CHEN C.C., HSIEH I.C. Celiac artery dissection presenting with abdominal and chest pain. *Am. J. Emerg. Med.*, 2010, **28** : 111 e113-115.
32. OZAKI N., WAKITA N., YAMADA A., TANAKA Y. Spontaneous dissection of the splanchnic arteries. *Interact. Cardiovasc. Thorac. Surg.*, 2010, **10** : 656-658.
33. ZEINA A.R., NACHTIGAL A., TROITSA A., ADMON G., AVSHOVICH N. Isolated spontaneous dissection of the celiac trunk in a patient with bicuspid aortic valve. *Vasc. Health Risk Manag.*, 2011, 6 : 383-386.
34. OZTURK T.C., YAYLACI S., YESIL O., CEVIK SE., GUNEYSEL O. Spontaneous isolated celiac artery dissection. *J. Res. Med. Sci.*, 2011, **16** : 699-702.
35. ZHANG T., ZHANG X., ZHANG X., JIANG J., ZHOU B. Endovascular treatment of isolated spontaneous celiac artery dissection. *Vascular*, 2012, **20** : 118-120.