

A rare cause of abdominal pain: Celiac compression syndrome

Altuğ Şenol*, Mert Köroğlu**, Mete Akın*, Yıldırım Songür*, Cem Koçkar*, Mehmet İşler*

*Süleyman Demirel Üniversitesi, Tıp Fakültesi, İç Hastalıkları A.D., Gastroenteroloji B.D., Isparta.

**Süleyman Demirel Üniversitesi, Tıp Fakültesi, Radyoloji Anabilim Dalı, Isparta.

Özet

Karın ağrısının nadir bir nedeni: Çölyak bası sendromu

Mezenterik iskemi karın ağrısının nadir bir nedenidir. Çölyak aks kompresyon sendromu, çölyak trunkusun genellikle median arkuat ligament tarafından eksternal kompresyona uğraması sonucu görülen oldukça nadir bir mezenterik iskemi sebebidir. Bu sendromun başlıca klinik bulguları, özellikle yemek sonrası olan karın ağrısı, diyare ve kilo kaybıdır. Tanı genellikle anjiyografi bulgularına dayanmaktadır. Renkli doppler ultrasonografi de tanıda yardımcı olabilir. Tedavide perkütan transluminal anjioplasti ve cerrahi yaklaşımlar önerilmektedir. Bu yazıda açıklanamayan karın ağrısı nedeniyle değerlendirilen ve çölyak aks kompresyon sendromu tanısı konulan 23 yaşında bir bayan hasta bildirilmiştir.

Anahtar kelimeler: Karın ağrısı, mezenterik iskemi, çölyak aks bası sendromu

Abstract

Mesenteric ischemia is an uncommon etiology of abdominal pain. Celiac axis compression syndrome (CACS) is an extremely rare cause of mesenteric ischemia due to external compression of the celiac trunk usually by median arcuat ligament. The clinical features of this syndrome includes abdominal pain, especially postprandial, diarrhea and weight loss. The diagnosis is usually depends on angiography findings. Color Doppler sonography can be helpful for diagnosis. Percutan transluminal angioplasty (PTA) and surgical approaches have been suggested for the treatment of CACS. We report here, a case of CACS in a 23 year old woman presenting with unexplained abdominal pain.

Key words: Abdominal pain, mesenteric ischemia, celiac axis compression syndrome

Introduction

Abdominal pain is a very common and troublesome clinical problem. The diagnosis is usually difficult because of different etiologic factors. It may be caused by a functional problem like gastroenteritis, or organic problems such as malignancy, bowel obstruction and mesenteric ischemia (1). Mesenteric ischemia is a severe clinical disorder due to decreased arterial blood supply to the intestine. The clinical characteristic feature of mesenteric ischemia is abdominal pain, especially postprandial, and it may be acute or chronic (2).

Celiac axis compression syndrome (CACS) is an extremely rare cause of mesenteric ischemia. This syndrome is an extreme condition of mesenteric ischemia due to external compression of the celiac trunk. CACS was first described by Harjola in a

young woman with chronic abdominal pain that he attributed to mesenteric ischemia caused by extrinsic compression of the celiac artery (3, 4). We report here, a case of CACS in a 23 year old woman presenting with unexplained abdominal pain.

Case Report

23 year old women was admitted to gastroenterology clinic because of continuous abdominal pain, which was began at around umbilicus and disseminate to abdomen and unrelated to meals. She had a history of pain with the same character for 3 years. In the past, she had a prior cystectomy, because of ovarian cyst. Approximately six months after from this first operation, she was admitted to general surgery clinic because of the same abdominal pain and segmentary bowel resection was performed with diagnosis of bridge ileus. Pathology result of resection material of bowel was reported as transmural infarctus. Because of persistent abdominal pain, patient was admitted

Corresponding Address: Arş. Gör. Dr. Mete Akın
Süleyman Demirel Üniversitesi, Tıp Fakültesi, İç Hastalıkları A.D.,
Gastroenteroloji B.D., Isparta.
Tel: 0505 5732010 - 0246 2112878
E-posta: drmeteakin@hotmail.com

Müracaat tarihi: 13.11.2009
Kabul tarihi: 16.03.2010

to general surgery clinic for the second time and laparoscopy was performed. Bridectomy and appendectomy was done for excessive brids. Colesistectomy was performed owing to persistent abdominal pain, approximately three weeks after from this operation. However, her symptoms persisted. When she was admitted to our clinic, on physical examination her abdomen was soft with mild excessive tenderness. Abdomen ultrasonography was normal. On laboratory tests; Hemoglobin :8,2 g/dl (12-16), platelet count:119,000/mm³ (130.000-400.000), leukocyte count :3,200/mm³ (4.800-10.800), AST: 44 U/L (0-37), ALT: 12 U/L (0-40), ALP: 42 U/L (10-100), erythrocyte sedimentation rate: 5 mm/hour, CRP and urine test were normal. The following tests were either normal, negative or nonreactive: iron indices, B12, folic acid, hepatitis A, B, C serologies, human immunodeficiency virus, thyroid function tests, serum protein and immunoelectrophoresis, FMF mutation test, aminolevulonic acid (ALA) and copro bilinogen levels. Esophagogastroduodenoscopy showed antral gastritis and colonoscopy was normal. We determined greater than 4-5-fold acceleration of flow in the celiac artery compared to the abdominal aorta on abdominal color Doppler ultrasonography (figure1).

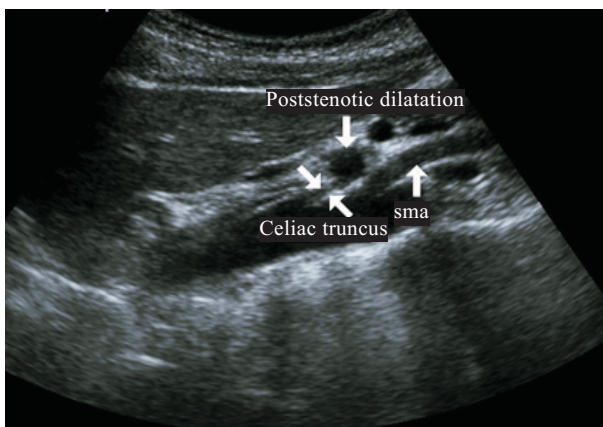


Figure 1: Stenosis at proximal portion of celiac artery and dilatation of poststenotic segment (arrows). SMA: Superior mesenteric artery

Due to this findings, CACS was considered and abdominal aortography, celiac axis and ve superior mesenteric artery angiogram was performed. Angiography showed %80 stenosis at the proximal portion of celiac artery and external compression especially from anterior and dilatation of poststenotic segment (figure 2). However, splenic artery and common hepatic artery showed good filling, arteria

hepatica propria, gastroduodenal artery, right and left hepatic artery showed little and low density filling, after celiac axis injection. Median arcuate ligament syndrome was considered and surgical intervention was predicted.

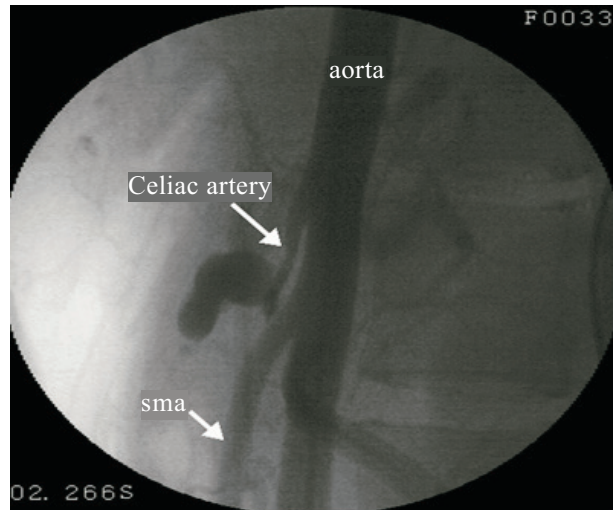


Figure 2: %80 stenosis at proximal portion of celiac artery and dilatation of poststenotic segment (big arrow). SMA: Superior mesenteric artery

Discussion

Celiac Axis Compression Syndrome (CACS) was first described in the 1960s but was not well understood until recently. CACS is a rare condition associated with the potential clinical presentations of upper abdominal pain, weight loss, hyperemesis, and the presence of epigastric bruits caused by inadequate blood flow through the celiac artery with resultant ischemia (5). In most cases, the external compression of celiac axis is by median arcuate ligament or celiac ganglion (6). Rarely it is caused by type B aortic dissection (7). In our case, celiac axis was copressed by the median arcuate ligament. The celiac trunk emerges from the aorta at the level of the aortic hiatus of the diaphragm, and the median arcuate ligament is formed by the fusion of the crura on either side of the aortic hiatus (8).

The symptoms can be variable. Most patients complain of abdominal postprandial pain which is usually epigastric. However, in some patients this condition has no relation to meals. Therefore, diagnosis was made after exclusion of other causes of abdominal pain. In our patient, pain was continuous and unrelated to meals. We suspected ischemic bowel disease (mesenteric ischemia), due to pathology result of intestinal resection material was reported as

transmural infarction and other causes of abdominal pain were excluded. Angiography is usually performed in the evaluation of patients with suspected mesenteric ischemia. Doppler ultrasound scanning has been used as a noninvasive technique for anatomic and physiologic assessment of the celiac artery (8, 9, 10). However, because of the high dependence on the performer's technique, the role of color Doppler sonography for the diagnosis of CACS is still controversial. Recently, in a study which was evaluated clinical and color Doppler sonographic features in a series of 59 cases, reported that, diagnosis of CACS was made if a greater than 2-fold acceleration of peak systolic flow in the celiac artery compared to the abdominal aorta (11). In our patient, first we performed portal vein color Doppler ultrasonography for diagnosis and greater than 4-5-fold acceleration of flow in the celiac artery compared to the abdominal aorta, stenosis at proximal portion of celiac artery and dilatation of poststenotic segment was detected (Figure 1). Due to this findings, we considered CACS and angiography was performed. Angiography demonstrated that approximately %80 stenosis at proximal portion of celiac artery, external compression especially from anterior and dilatation of poststenotic segment (Figure 2). Multi-detector CT angiography has become a more favorable modality in diagnosing mesenteric ischemia because it is rapid, non-invasive and less expensive, but we didn't have multi-detector CT in our hospital. In the treatment of CACS, percutan transluminal angioplasty (PTA) and surgical approaches were suggested in the documents reviewed (12, 13, 14). Although in a few cases reported the symptoms were completely relieved after PTA, the symptoms of most patients receiving PTA are only relieved for a short time period (15). Surgical approaches for the CACS include, division of the median arcuate ligament or arterial reconstruction or by-pass (16). In our patient, surgical intervention was predicted for treatment of CACS.

In the evaluation of prognosis, in a large study which include the long-term evaluation of CACS patients treated by surgery, reported that, %83 of patients were asymptomatic in the first 6 months after decompression, but only %41 of patients remained asymptomatic in a follow-up period ranging from 3 to 11 years (13).

Conclusion

Eventually, diagnosis of patient with abdominal pain couldn't be made easily. Sometimes, unnecessary operations were performed. This condition is related to mortality, morbidity and undesirable cost. Therefore, mesenteric ischemia and CACS as a rare cause of abdominal pain, it should be considered in patients presenting with especially postprandial and unexplained abdominal pain.

References

1. Clouse RE, Mayer EA, Aziz Q, et al. Functional abdominal pain syndrome. *Gastroenterology* 130: 1492-1497, 2006.
2. Martinez JP, Hogan GJ. Mesenteric ischemia. *Emerg Med Clin N Am* 22: 909-928, 2004.
3. Hariola PT. A rare obstruction of the coeliac artery. *Annals Chirurgiae et Gynaecologiae Fenniae* 1963;52:547-9.
4. Jamieson C.W. Coeliac axis compression syndrome *British Medical Journal* Volume .1986. 293:159-160
5. Loukas M, Pinyard J, Vaid S, Kinsella C, Tariq A. Clinical Anatomy of Celiac Artery Compression Syndrome:A Review . *Clinical Anatomy* 20:612-617 (2007).
6. Bech FR. Celiac artery compression syndromes. *Surg Clin N Am* 77: 409-424, 1997.
7. Fujisawa Y, Morishita K, Fukada J, Hachiro Y, Kawaharada N, Abe T. Celiac artery compression syndrome due to acute type B aortic dissection. *Ann Vasc Surg* 19: 553-556, 2005.
8. Alehan D, Dogan ÖF. A Rare Case: Celiac Artery Compression Syndrome in an Asymptomatic Child. *Journal of Pediatric Surgery*, Vol 39, No 4 (April), 2004: 645-647.
9. Moneta GL, Lee RW, Yeager RA, et al: Mesenteric duplex scanning: A blinded prospective study. *J Vasc Surg* 17:79-87, 1993
10. Roobottom CA, Dubbins PA: Significant disease of the celiac mesenteric arteries in asymptomatic patients: Predictive value of Doppler sonography. *AJR* 161:985-988, 1993)
11. 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* 25:299-305
12. Carbonell AM, Kercher KW, Heniford BT, Mhashelkar Y, Talekar K. Laparoscopic management of median arcuate ligament syndrome. *Surg Endosc* 19: 729, 2005.
13. Evans WE. Long-term evaluation of the celiac band syndrome. *Surgery* 76: 867-871, 1974.
14. Yamaguchi H, Arita Y, Ohkubo A, et al. A case of celiac axis compression syndrome relieved by

- percutaneous transcatheter angioplasty. *Nippon Shokakibyō Gakkai Zasshi (The Japanese Journal of Gastroenterology)* 89: 528-531, 1992 (in Japanese).
15. Otte JA, Geelkerken RH, Oostveen E, Mensink PB, Huisman AB, Kolkman JJ. Clinical impact of gastric exercise tonometry on diagnosis and management of chronic gastrointestinal ischemia. *Clin Gastroenterol Hepatol* 3: 660-666, 2005.
 16. Kokotsakis J.N, Lambidis C.D, Lioulis A.G, Skouteli E.T, Bastounis E.A, Livesay J.J. Celiac artery compression syndrome. *Cardiovascular Surgery*. 2000; 8: 3: 219-222.