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CASE REPORT

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A 56-year-old female with celiac artery compression syndrome recovering through dietary changes and weight gain

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Abstract

Celiac artery compression syndrome is a rare condition characterized by extrinsic compression of the celiac artery by the median arcuate ligament, which leads to chronic, recurrent postprandial epigastric pain. Although traditional treatment of this syndrome involves laparotomy to release the median arcuate ligament, here we report on the rare case of a middle-aged woman recovering with no specific treatment through her use of a food diary to change her diet which resulted in gradual weight gain. This suggests that in some cases, a conservative therapy for celiac artery compression syndrome should be tried before choosing surgical intervention.

KEYWORDS

angiography, celiac artery compression syndrome, food diary, postprandial epigastric pain

1 | INTRODUCTION

Celiac artery compression syndrome is defined as chronic, recurrent abdominal pain related to compression of the celiac artery by the median arcuate ligament.[1-3]

We experienced the rare case of a middle-aged female who was diagnosed with celiac artery compression syndrome who recovered with no specific treatment other than gaining weight by changing her dietary habits.

2 | CASE REPORT

A 56-year-old female patient was evaluated for a 3-week history of postprandial epigastric pain. The pain radiated to her back, sometimes with a cold sweat. Those symptoms started 2 hours after taking food, mostly after dinner, and continued for 2-3 hours. She had no appreciable past medical history and took no medicine. She was a nonsmoker and drank a can of beer most days of the week. She lost 6 kg over the previous 4 years although she explained the weight loss was from the stress of her new career and not from the postprandial pain.

On examination, her height was 146.4 cm, body weight 42.9 kg, BMI 20.02 kg/m^2, blood pressure 150/90 mmHg, heart rate 64 beats per minute and regular, body temperature 36.1°C and respiratory rate 18 breaths per minute. There was no sign of anemia or jaundice in her conjunctiva. Her abdomen was soft with slight tenderness around her subumbilical area. A subtle epigastric bruit was detected on careful auscultation. The remainder of the examination was normal.

Her routine laboratory studies were all normal. A plain CT of her abdomen showed no particular findings except for a left ovarian cyst. The gynecologist performed a transvaginal ultrasonography that revealed no relationship between this cyst and the symptoms.

We advised her to keep a daily food diary after meals and record her symptoms to examine potential relationships between them. As a result, we discovered she experienced the same type of postprandial epigastric pain four times over the next fortnight. Each symptom started 2-4 hours after dinner or lunch and continued for 2-4 hours. She ate various types of foods, and there were no specific foods or beverages (including alcoholic) that related to those symptoms. We suspected those symptoms were due to mesenteric vascular disease rather than food allergies.

We performed contrast-enhanced CT and suspected stenosis on her origin of the celiac artery (Figure 1). Therefore, we consulted a

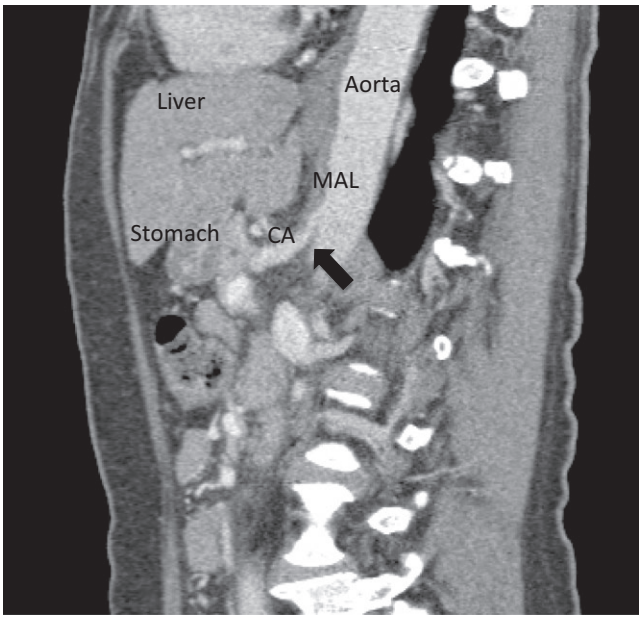

FIGURE 1 Contrast-enhanced CT indicated stenosis (arrow) on her origin of the celiac artery (CA) due to compression by the median arcuate ligament (MAL)

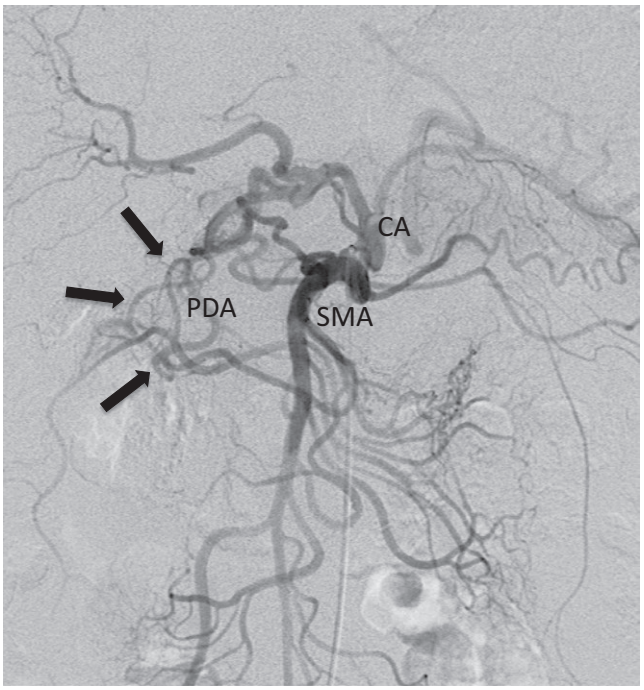

FIGURE 2 Retrograde filling of the celiac artery (CA) from the superior mesenteric artery (SMA) through a well-developed pancreaticoduodenal arcade (PDA) (arrows)

radiologist who performed an angiography to investigate the condition of her celiac artery in detail. The result of her angiography revealed a stenosis of the origin of the celiac artery due to the median arcuate ligament indenting upon the celiac trunk and causing downward angulation. Additional findings included retrograde filling of the celiac axis from the superior mesenteric artery through a well-developed pancreaticoduodenal arcade (Figure 2). Stasis of the hepatic artery was seen on the angiography due to this retrograde filling. The range of retrograde filling of the celiac artery became wider during the expiratory phase than during the inspiratory phase, suggesting the stenosis became more severe during expiration. These findings were compatible with celiac artery compression syndrome.

We explained the mechanism of this syndrome and presented the option of surgical treatment to her, but she refused. Therefore, we started conservative management to monitor progression of the symptoms. Through her diary she found that taking less food at dinner and increasing the number of meals lessened the severity of the postprandial symptoms. Two months later, she gained back her former weight, to around 45 kg, and no symptoms have occurred since.

3 | DISCUSSION

Lipshutz first described celiac artery compression from autopsy studies in 1917, followed by Harjora who first described a case as a clinical syndrome with postprandial abdominal pain and epigastric bruit in 1967.[1-3] The incidence rate of the celiac artery compression due to the median arcuate ligament is 10-24% of the healthy portion of the Western population.[4] Meanwhile, two lower incidence rates of 2.3% and 7.3% have been reported for Japan and Korea, respectively.[5-7] This syndrome usually occurs in young, thin females.[3] Indeed, Jen-Wei et al.[5] reported a mean age and BMI of 28.4 years and 18.2 kg/m^2, respectively, from 14 cases in Taiwan, 10 of whom were female (71%). Our case, in contrast, was a middle-aged short female which indicates this syndrome can occur among multiple age groups and not only among females with low BMIs.

Although the triad of postprandial epigastric pain, unintentional weights loss and/or abdominal bruit is a classical manifestation of celiac artery compression syndrome, the true clinical features of this syndrome are variable[5,8] and often diagnosed through exclusion.[3]

The mechanism of this pain is not completely understood. The most accepted theory is that the pain is from the mesenteric ischemia through the steal phenomenon, namely postprandial steal via collaterals from the superior mesenteric artery.[5] Another theory suggests that the pain is caused by chronic irritation of the celiac plexus with subsequent splanchnic vasoconstriction and ischemia.[3]

For diagnosis, the lateral view of aortography had been thought a gold standard.[2,3,5] CT angiography is also considered useful recently, as it reveals information about the relationship of the celiac artery with other structures including the diaphragm.[2-4] Diagnosis remains a challenge, however, and imaging findings must be correlated with the patient's clinical history.[1]

Various surgical approaches have been reported in the literature as the best option for treatment.[2,5] Although there are few case reports that referred for surgical procedures from East Asian countries, Jen-Wei et al.[5] reported from Taiwan that no successful cases were found in 12 conservative therapy patients and one patient of two who received surgical treatment recovered without recurrence. On the other

hand, there was a case from Japan whose symptoms were relieved through conservative management for 2.5 years with pharmacological therapy to facilitate gastrointestinal motility.[9] Our case, in addition, recovered by reviewing her diet through her food diary and gaining weight without pharmacological therapy or surgical intervention. These findings indicate to us that surgery is not the only means of treatment and that a wait and see attitude could be a viable alternative in some cases. There were no articles, as far as we could find, that refer to the relationship between symptom relief and weight gain in patients with celiac artery compression syndrome. Further investigations are required to reveal the true principles of its clinical course.

4 | CONCLUSION

We presented a rare case of celiac artery compression syndrome in which the patient recovered through dietary changes and weight gain. Our case suggests that in some cases, a conservative approach can be a treatment choice for celiac artery compression syndrome.

CONFLICT OF INTEREST

The authors have stated explicitly that there are no conflicts of interest in connection with this article.

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