

e-ISSN 1941-5923 © Am J Case Rep. 2025: 26: e946075 DOI: 10.12659/AJCR.946075

Accepted: 2025.01.04 Available online: 2025.01.23 Published: 2025.03.06

Chronic Post-Prandial Epigastric Pain Associated with Median Arcuate Ligament Syndrome and Atherosclerosis of the Celiac Trunk in An Elderly Woman: A Case Report

Authors' Contribution: Study Design A Data Collection B Statistical Analysis C Data Interpretation D Manuscript Preparation E Literature Search F Funds Collection G

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None declared Conflict of interest: None declared

Patient:

Female, 66-year-old

Final Diagnosis:

Median arcuate ligament syndrome and atherosclerosis of the celiac trunk

Symptoms:

Chronic post-prandial epigastric pain

Clinical Procedure:

Cardiology • Gastroenterology and Hepatology

Objective:

Specialty:

Unusual clinical course

Background:

Post-prandial abdominal pain due to bowel ischemia can be caused by stenosis (atherosclerosis) or by compression of the arteries of the celiac axis. Median arcuate ligament syndrome (MALS) results from compression at the origin of the celiac trunk by the arcuate ligament. This report describes a 66-year-old woman with chronic post-prandial epigastric pain associated with atherosclerosis of the celiac trunk, managed with angioplasty and stenting combined with MALS.

Case Report:

A 66-year-old female patient with a history of dyslipidemia presented with chronic epigastric pain with postprandial episodes for 4 years. Two years before the admission, her pain increased with meals and was not relieved by empirical treatment for gastritis. An esophagogastroduodenoscopy and colonoscopy showed chronic gastritis. One year later, a resection of the gastric submucosal tumor was performed, without improving her symptoms. In this presentation, the prompt computed tomography revealed hook-shaped stenosis of the celiac trunk and mild post-stenosis dilatation, highly suggestive of MALS. However, the patient refused to undergo surgery despite the benefit of this intervention. Concurrently, moderately severe atherosclerosis of the celiac trunk was detected during intra-vessel imaging. The patient was treated individually and underwent angioplasty with stenting. At a 5-month follow-up, the patient's condition was stable and she had no gastrointestinal symptoms.

Conclusions:

Due to the low prevalence and nonspecific symptoms of MALS, physicians should be highly suspicious of this disease, especially in patients with post-prandial abdominal pain. Angioplasty and stenting can be performed in selected patients with MALS and atherosclerosis of the celiac trunk.

Keywords:

Angioplasty • Atherosclerosis • Case Reports • Celiac Artery • Ligaments

Full-text PDF:

https://www.amjcaserep.com/abstract/index/idArt/946075









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Introduction

The median arcuate ligament is an anatomical structure located in the upper abdomen. It is a tendinous portion of the diaphragm that arches over the aorta and forms the aortic hiatus, typically at the level of the T12 vertebra, just above the origin of the celiac trunk [1]. In some individuals, the median arcuate ligament is positioned lower than normal and compresses the celiac trunk, leading to median arcuate ligament syndrome (MALS) [2]. The syndrome is a relatively uncommon condition, with an estimated prevalence of approximately 2 cases per 100 000 individuals. MALS typically affects young women, with a female-to-male ratio of 2: 1 to 3: 1, and predominantly occurs in individuals within the age range of 20 to 40 years [3]. The underlying reasons for the observed predominance in women remain uncertain; however, it may be related to a tendency for the celiac artery to originate at a more cephalad position in females than in males [4].

The exact cause of this anatomical malformation is unknown. Patients with MALS experience a wide range of nonspecific symptoms, including post-prandial epigastric pain, nausea, vomiting, food fear, and weight loss [5]. The diagnosis of MALS is a diagnosis of exclusion supported by imaging methods, such as computed tomography (CT) and Doppler ultrasound [3]. Atherosclerosis should be excluded, because the celiac artery is the most frequently affected site of mesenteric artery atherosclerosis [6]. There is currently no consensus on the treatment of MALS. Treatment is mainly surgery to remove the median arcuate ligament (MAL), which relieves compression on the celiac trunk and plexus [1]. Celiac artery revascularization is generally reserved for refractory cases if symptoms persist after surgery [3]. Several cases of MALS have been reported, often presenting diagnostic challenges [7]. However, to the best of our knowledge, there has been no documented case involving concurrent MALS and celiac artery atherosclerosis. Furthermore, an approach to managing these coexisting conditions may remain unclear.

Herein, this report describes a 66-year-old woman with chronic post-prandial epigastric pain associated with atherosclerosis of the celiac trunk, managed with angioplasty and stenting, combined with MALS.

Case Report

A 66-year-old female patient with a history of uncontrolled dyslipidemia presented to the gastrointestinal clinic with chronic abdominal pain for 4 years. Four years prior to this presentation, the patient reported experiencing mildly dull epigastric pain that typically occurred after meals, lasting for 1 to 2 h, accompanied by mild abdominal distension. The symptoms

were initially minimal, occurring 1 to 3 times per month. The pain increased in severity over the years. Two years later, it worsened with large meals, and the symptoms gradually became more frequent and severe, occurring almost daily, leading the patient to restrict food intake and divide meals into smaller portions. The patient also sometimes experienced nausea; however, she denied weight loss, diarrhea, and fever. An esophagogastroduodenoscopy (EGD) and colonoscopy showed no significant abnormalities. The patient was empirically treated for gastritis and irritable bowel disease; however, symptoms still persisted. A year before the presentation, the patient underwent a resection of a gastric submucosal tumor, with benign pathology. The patient's symptoms did not improve significantly after the tumor removal, prompting her to seek our hospital.

At initial evaluation, the patient was in no distress, and the physical examination revealed only mild epigastric tenderness, with no bruit or other significant findings. Laboratory test results revealed no significant findings except for dyslipidemia (low-density lipoprotein cholesterol: 4.9 mmol/L). The abdominal ultrasound did not suggest any possible causes. A repeat EGD showed chronic gastritis, which did not correlate with the patient's clinical scenario. Subsequently, an abdominal CT scan with contrast was performed to investigate other potential causes of the pain. The results revealed hook-shaped stenosis of the celiac trunk at the upper margin of the L1 vertebra and mild post-stenosis dilatation, highly suggestive of MALS (Figure 1). The patient exhibited a strong preference against surgical intervention, including minimally invasive laparoscopic surgery, due to concerns regarding the associated risks of surgery, despite discussions regarding the potential benefits of decompression intervention.

Due to the patient's advanced age and the presence of dyslipidemia, atherosclerosis was suspected to play a role in pathogenesis. A discussion between the patient, her family, and a team of physicians, including a gastroenterologist and a cardiovascular interventionist, was held to discuss the risks and benefits of each intervention, including surgery and angioplasty, leading to the agreement to perform endovascular angioplasty with stent implantation. Digital subtraction angiography confirmed the nearly total occlusion of the celiac trunk, with the position suggesting MALS (Figure 2). During the procedure with a 5×18 mm Herculink Elite (Abbott Vascular, Santa Clara, CA, USA) stent, intravascular ultrasound revealed concurrent moderately severe intra-luminal atherosclerotic stenosis of approximately 60% to 70% (Figure 3). Post-implantation angiography revealed a significant improvement in blood flow to the branches of the celiac trunk (Figure 4).

After the intervention, the patient remained in stable condition but had mild urticaria. A follow-up abdominal CT showed

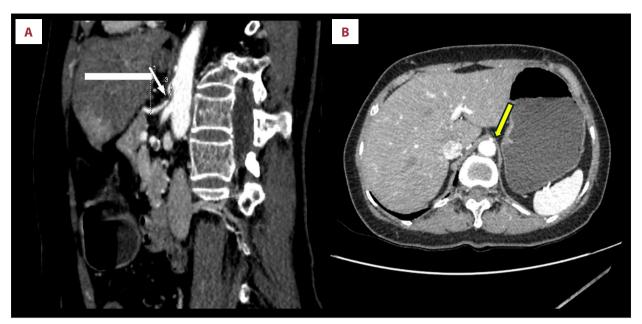


Figure 1. Computed tomography scans with contrast. (A) Image shows hook-shaped stenosis of the origin of the celiac trunk at the upper margin of L1 (white arrow) and mildly post-stenosis dilatation (sagittal view); (B) a low-density structure arch (a fibrous band that joins the 2 crura of the diaphragm) over the origin of the celiac trunk (coronal view) is marked with a yellow arrow.

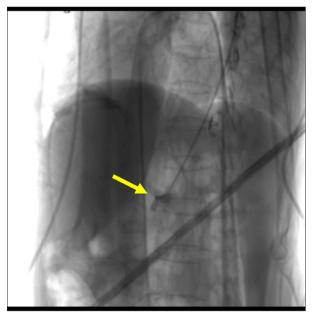


Figure 2. Digital subtraction angiography of the celiac trunk confirmed nearly total occlusion of the celiac trunk (yellow arrow shows no contrast after the celiac trunk).

a well-placed stent with good flow inside and a minor curve at the position of the ligament. The patient reported remarkable improvement in her epigastric pain and was discharged on day 5 of admission with a regimen that included low-dose aspirin, clopidogrel, rosuvastatin, and bisoprolol. At the 5-month follow-up, the patient was in stable condition and noted that

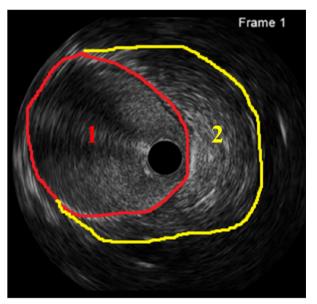


Figure 3. Intravascular ultrasound of the celiac trunk shows moderately severe concurrent intra-luminal atherosclerotic stenosis (1: the lumen of the celiac trunk, 2: atherosclerosis).

she could consume larger portions of meals without experiencing discomfort or digestive issues. The patient will be reassessed every 3 months, and a CT scan will be performed after 6 months or if her symptoms recur.

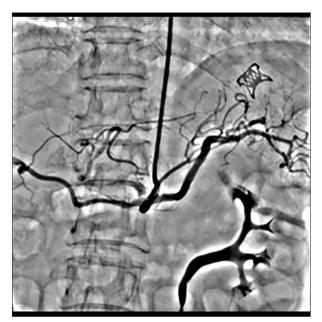


Figure 4. Digital subtraction angiography of the celiac trunk after stenting shows a significant improvement in blood flow of celiac branches after angioplasty with stenting.

Discussion

To the best of our knowledge, this is the first case presenting chronic post-prandial epigastric pain associated with MALS and celiac artery atherosclerosis. Due to the low prevalence and nonspecific symptoms of MALS, it could be misdiagnosed. This case made physicians highly suspicious of this disease, especially in patients with post-prandial abdominal pain. Furthermore, angioplasty and stenting can be considered in selected patients with MALS and atherosclerosis of the celiac artery.

Most patients with MALS are asymptomatic, and the high recurrence rate after decompression illustrates the complexity of its pathophysiology, suggesting that it involves more than just ischemia [5]. As seen in the present case, finding another pathological process involved, such as atherosclerosis, is essential. The pathophysiological process of MALS includes ischemia to the organs supplied by the celiac trunk, with or without disturbances in the neural transmission of the celiac plexus [3]. The compression of the celiac trunk can reduce the blood supply to the foregut and foregut-derived organs, including the distal esophagus, stomach, proximal duodenum, liver, pancreas, gallbladder, and spleen [8]. Additionally, the pressure exerted on the nearby celiac plexus can disrupt the transmission of neural impulses from and to these organs, possibly playing a role in the pathophysiology of MALS. However, chronic compression could predispose to other secondary pathologic processes, especially atherosclerosis, which can worsen arterial constriction [9]. In one study with 120 unselected Finnish autopsy patients, 29% of patients had stenosis of mesenteric arteries, with the celiac artery being the most common site [6]. Furthermore, recent findings by Emre et al provided no evidence to support the hypothesis that a superiorly positioned celiac trunk contributes to MALS. The distance between the celiac trunk and the superior mesenteric artery was measured at a relatively shorter distance of 9.19 mm, compared with that in the existing literature [10].

Symptoms of MALS are nonspecific and various range. The most common manifestation is chronic abdominal pain, usually located in the epigastrium and occurring post-prandially. Additionally, patients can experience unintentional weight loss, nausea, vomiting, and diarrhea [2]. Physical examination is usually insignificant, but weight loss and epigastric tenderness can be present. An epigastric bruit may be heard on auscultation up to 35%, particularly during expiration when the median arcuate ligament moves downward [11]. Many patients with MALS are asymptomatic because of the collateral circulation that supplied the foregut, while others can present lifethreatening hemorrhage due to post-stenosis aneurysm rupture [12]. Compared with other reported cases, our case shares similarities with 3 others regarding diagnostic challenges and initial misdiagnosis [7]. However, our case did not present with weight loss or vomiting, which are part of the classic symptom triad (post-prandial epigastric pain, weight loss, nausea, and vomiting). This may be attributed to the chronic nature of the 4-year clinical course and differences in dietary patterns, compared with the 3-month symptom history observed in the other reports [7]. Therefore, this syndrome should be considered in the differential list when patients present with chronic abdominal pain, especially when occurring post-prandially and when symptoms are not relieved with empiric treatment, which guides the prompt imaging methods.

The diagnosis of MALS typically involves evidence of celiac trunk stenosis and excludes other causes of abdominal pain [2]. EGD and CT scans could help rule out more common conditions. Evidence of celiac trunk stenosis can be gained with duplex ultrasound, CT angiography, or digital subtraction angiography [3]. Duplex ultrasound is considered an initial investigation, because of cost-effectiveness, noninvasiveness, and lack of radiation exposure. Gruber et al highlighted that specific ultrasound findings, such as a celiac trunk deflection angle greater than 50° and an expiratory peak systolic velocity over 350 cm/s, can provide high sensitivity and specificity for MALS diagnosis [13]. Classical CT findings of MALS include proximal celiac trunk narrowing with a characteristic hooked configuration (a visual fold angle < 135°) with post-stenotic dilatation, best viewed on sagittal images, which may differentiate it from atherosclerosis [14,15]. However, these findings could not exclude the atherosclerosis of the celiac trunk. In our case, we did not perform duplex ultrasound, due to a

thickening abdominal wall, resulting in a bad window for ultrasound. However, CT angiography revealed typical findings for MALS, similar to other reported cases [7]. Therefore, imaging modalities, including Doppler ultrasound and CT scans, are essential for confirming the diagnosis [3].

Decompression or intervention is recommended only when symptomatic MALS is confirmed, and other more common causes of symptoms are excluded [16]. The primary goal of MALS management should be to intervene in the pathophysiological process, which requires individual investigation and tailored approaches [5]. Currently, most patients with MALS are treated with laparoscopic MAL release. Symptomatic resolution is usually achieved within 6 weeks after the procedure [11]. The initial success rate ranges from 93% to 94%, and revascularization can further increase the success rate [3]. However, the symptomatic recurrence rate is also high, at 38% for laparoscopic decompression and 50% for open decompression [11]. Furthermore, several complications of both the open and laparoscopic surgery were recorded, such as pneumothorax, celiac artery bleeding, aortic puncture, and splenic infarction [17]. Due to the high recurrence rate and unclear associated factors of MALS, a recent study showed that hybrid laparoscopic and endovascular treatment during the same procedure had better outcomes, with no recurrent symptoms within a 6-month follow-up [1].

Our patient's case was quite challenging in terms of diagnosis and treatment. Due to aging and dyslipidemia, our patient had risk factors for atherosclerotic disease. Digital subtraction angiography and intravascular ultrasound confirmed the presence of moderately severe atherosclerosis of the celiac trunk, which may explain the progressive symptoms of this patient

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1 year earlier. When stenosis is caused not only by external compression from the ligament but also by atherosclerosis, the optimal management approach remains controversial. Apparently, ligament incision alone may not fully resolve the stenosis in such cases. Angioplasty is significantly less invasive and could help relieve internal stenosis; however, it alone carries a higher risk of treatment failure and re-stenosis and requires long-term antiplatelet therapy [1]. Therefore, a combination of laparoscopic release of the median arcuate ligament and angioplasty with stenting may have been the most appropriate approach for this patient. However, the patient opted to defer invasive procedures and surgery, which resulted in endovascular angioplasty and stent implantation as the alternative treatment. The patient was in stable condition at a 5-month follow-up, and a longer monitor is needed to observe the long-term prognosis and guide for further treatment.

Conclusions

In conclusion, median arcuate ligament syndrome is a rare condition with diverse presentations. Physicians should raise high suspicion of this disease in patients with chronic post-prandial epigastric pain. Imaging studies such as duplex ultrasound and CT angiography are crucial in confirming the diagnosis. Angioplasty and stenting can be performed in selected patients with MALS and atherosclerosis of the celiac trunk.

Declaration of Figures' Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

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