

# World Journal of *Clinical Cases*

*World J Clin Cases* 2022 February 26; 10(6): 1754-2052



**OPINION REVIEW**

- 1754 Gut-brain axis: Focus on gut metabolites short-chain fatty acids  
*Guo C, Huo YJ, Li Y, Han Y, Zhou D*

**MINIREVIEWS**

- 1764 Association between direct-acting antiviral agents in hepatitis C virus treatment and hepatocellular carcinoma occurrence and recurrence: The endless debate  
*Kamal A, Elsheaita A, Abdelnabi M*

**ORIGINAL ARTICLE****Retrospective Cohort Study**

- 1775 Effects of bilirubin on perioperative myocardial infarction and its long-term prognosis in patients undergoing percutaneous coronary intervention  
*Li Y, Li DB, Zhao LD, Lv QB, Wang Y, Ren YF, Zhang WB*
- 1787 Disease exacerbation is common in inflammatory bowel disease patients treated with immune checkpoint inhibitors for malignancy  
*Rubin SJS, Balabanis T, Gubatan J, Habtezion A*
- 1795 Multidrug-resistant organisms in intensive care units and logistic analysis of risk factors  
*Han Y, Zhang J, Zhang HZ, Zhang XY, Wang YM*

**Retrospective Study**

- 1806 Change and impact of left ventricular global longitudinal strain during transcatheter aortic valve implantation  
*Zhang H, Xie JJ, Li RJ, Wang YL, Niu BR, Song L, Li J, Yang Y*

**Observational Study**

- 1815 Early detection of noise-induced hearing loss  
*Meng ZL, Chen F, Zhao F, Gu HL, Zheng Y*
- 1826 Empathetic nursing with mindful cognitive therapy for fatigue, depression, and negative emotions in leukemia patients undergoing long-term chemotherapy  
*Lu YY, Lu XM, Shao CY, Wang CC, Xu TT, Zhang BL*

**Prospective Study**

- 1834 Superior pancreatic lymphadenectomy with portal vein priority *via* posterior common hepatic artery approach in laparoscopic radical gastrectomy  
*Zhang YJ, Xiang RC, Li J, Liu Y, Xie SM, An L, Li HL, Mai G*

**Randomized Controlled Trial**

- 1843 Systematic nursing interventions in gastric cancer: A randomized controlled study  
*He F, He RX*

**META-ANALYSIS**

- 1852 Impact of adding opioids to paravertebral blocks in breast cancer surgery patients: A systematic review and meta-analysis  
*Chen MH, Chen Z, Zhao D*

**CASE REPORT**

- 1863 Multiple different remote epidural hematomas after craniotomy: A case report  
*He Q, Tao CY, Fu RH, You C*
- 1869 Tuberculous pericarditis-a silent and challenging disease: A case report  
*Lucero OD, Bustos MM, Ariza Rodríguez DJ, Perez JC*
- 1876 Transileocolic endovascular treatment by a hybrid approach for severe acute portal vein thrombosis with bowel necrosis: Two case reports  
*Shirai S, Ueda T, Sugihara F, Yasui D, Saito H, Furuki H, Kim S, Yoshida H, Yokobori S, Hayashi H, Kumita SI*
- 1883 Efficacy of EGFR-TKI sequential therapy in patients with EGFR exon 19 insertion-positive non-small-cell lung cancer: A case report  
*Shan BB, Li Y, Zhao C, An XQ, Zhang QM*
- 1889 Novel compound heterozygous variants in the TAF6 gene in a patient with Alazami-Yuan syndrome: A case report  
*Lin SZ, Feng JH, Sun LP, Ma HW, Wang WQ, Li JY*
- 1896 Asymmetric limb weakness in Guillain-Barré syndrome: Three case reports  
*Hu M, Li X, Wong HY, Feng XG, Wang YZ, Zhang GR*
- 1903 Modified treatment of knee osteoarthritis complicated with femoral varus deformity: A case report  
*Xu SM, Li W, Zhang DB, Bi HY, Gu GS*
- 1909 Novel HNF1A gene mutation in maturity-onset diabetes of the young: A case report  
*Xu Q, Kan CX, Hou NN, Sun XD*
- 1914 Cerebral corridor creator for resection of trigone ventricular tumors: Two case reports  
*Liu XW, Lu WR, Zhang TY, Hou XS, Fa ZQ, Zhang SZ*
- 1922 Left abdominal wall proliferative myositis resection and patch repair: A case report  
*Xing RW, Nie HQ, Zhou XF, Zhang FF, Mou YH*
- 1929 Concurrent ankylosing spondylitis and myelodysplastic syndrome: A case report  
*Xu GH, Lin J, Chen WQ*

- 1937** Life-threatening subclavian artery bleeding following percutaneous coronary intervention with stent implantation: A case report and review of literature  
*Shi F, Zhang Y, Sun LX, Long S*
- 1946** Cryptogenic organizing pneumonia associated with pregnancy: A case report  
*Lee YJ, Kim YS*
- 1952** Eosinophilia complicated with venous thromboembolism: A case report  
*Su WQ, Fu YZ, Liu SY, Cao MJ, Xue YB, Suo FF, Liu WC*
- 1961** Neck and mediastinal hematoma caused by a foreign body in the esophagus with diagnostic difficulties: A case report  
*Wang LP, Zhou ZY, Huang XP, Bai YJ, Shi HX, Sheng D*
- 1966** Therapeutic endoscopy of a Dieulafoy lesion in a 10-year-old girl: A case report  
*Chen Y, Sun M, Teng X*
- 1973** Cavernous hemangioma of an intrapancreatic accessory spleen mimicking a pancreatic tumor: A case report  
*Huang JY, Yang R, Li JW, Lu Q, Luo Y*
- 1981** Surgery and antibiotics for the treatment of lupus nephritis with cerebral abscesses: A case report  
*Hu QD, Liao LS, Zhang Y, Zhang Q, Liu J*
- 1991** Median arcuate ligamentum syndrome: Four case reports  
*Kim JE, Rhee PL*
- 1998** Novel ABCB4 mutations in an infertile female with progressive familial intrahepatic cholestasis type 3: A case report  
*Liu TF, He JJ, Wang L, Zhang LY*
- 2007** Primary duodenal dedifferentiated liposarcoma: A case report and literature review  
*Kim NI, Lee JS, Choi C, Nam JH, Choi YD, Kim HJ, Kim SS*
- 2015** Implant site development using titanium plate and platelet-rich fibrin for congenitally missed maxillary lateral incisors: A case report  
*Zhang TS, Mudalal M, Ren SC, Zhou YM*
- 2023** Successful embolization of an intrahepatic portosystemic shunt using balloon-occluded retrograde transvenous obliteration: A case report  
*Saito H, Murata S, Sugihara F, Ueda T, Yasui D, Miki I, Hayashi H, Kumita SI*
- 2030** Bilateral pneumothorax and pneumomediastinum during colonoscopy in a patient with intestinal Behcet's disease: A case report  
*Mu T, Feng H*
- 2036** Acute kidney injury due to intravenous detergent poisoning: A case report  
*Park S, Ryu HS, Lee JK, Park SS, Kwon SJ, Hwang WM, Yun SR, Park MH, Park Y*

**2045** Vaginal enterocele after cystectomy: A case report

*Liu SH, Zhang YH, Niu HT, Tian DX, Qin F, Jiao W*

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**RESPONSIBLE EDITORS FOR THIS ISSUE**

Production Editor: *Ying-Yi Yuan*; Production Department Director: *Xu Guo*; Editorial Office Director: *Jin-Lei Wang*.

**NAME OF JOURNAL**

*World Journal of Clinical Cases*

**ISSN**

ISSN 2307-8960 (online)

**LAUNCH DATE**

April 16, 2013

**FREQUENCY**

Thrice Monthly

**EDITORS-IN-CHIEF**

Bao-Gan Peng, Jerzy Tadeusz Chudek, George Kontogeorgos, Maurizio Serati, Ja Hyeon Ku

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<https://www.wjgnet.com/2307-8960/editorialboard.htm>

**PUBLICATION DATE**

February 26, 2022

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**ARTICLE PROCESSING CHARGE**

<https://www.wjgnet.com/bpg/gerinfo/242>

**STEPS FOR SUBMITTING MANUSCRIPTS**

<https://www.wjgnet.com/bpg/GerInfo/239>

**ONLINE SUBMISSION**

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## Median arcuate ligamentum syndrome: Four case reports

Ji Eun Kim, Poong Lyul Rhee

**Specialty type:** Gastroenterology and hepatology

**Provenance and peer review:** Unsolicited article; Externally peer reviewed.

**Peer-review model:** Single blind

**Peer-review report's scientific quality classification**

Grade A (Excellent): 0  
Grade B (Very good): B  
Grade C (Good): C, C  
Grade D (Fair): 0  
Grade E (Poor): 0

**P-Reviewer:** Baryshnikova NV, Ghannam WM, Martínez-Pérez A

**Received:** August 23, 2021

**Peer-review started:** August 23, 2021

**First decision:** November 7, 2021

**Revised:** November 12, 2021

**Accepted:** January 19, 2022

**Article in press:** January 19, 2022

**Published online:** February 26, 2022



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### Abstract

#### BACKGROUND

Median arcuate ligamentum syndrome (MALS) is a disease entity with unclear pathogenesis. If it is not considered in advance, the clinical diagnosis of the disease is very difficult because patients complain of digestive discomfort including pain. However, this characteristic is not specific to MALS. There have been no studies to assist in making a quick diagnosis. The aim of this case series was to recognize that MALS must be considered as a differential factor in the cause of abdominal pain.

#### CASE SUMMARY

We described cases in which four patients complained of abdominal pain over a long period but in whom a diagnosis of MALS could not be made. If the gastroenterologist does not take into account abdominal pain in advance, the patient is considered an asymptomatic gallstone patient and has their gallbladder removed despite imaging evaluation. The patient may also be considered a psychiatric patient and may be administered psychiatric drugs over a long period. In all four cases in this report, the patients experienced abdominal pain. In three cases, the diagnosis was possible by the clinician's judgment considering both clinical symptoms and imaging techniques shortly after the onset of symptoms. However, in one case that lasted over 20 years, a clear diagnosis was not possible. Even after complaining of colicky pain and performing a cholecystectomy, the diagnosis was made only after the symptoms persisted. In all four cases, the symptoms were relieved by neuromodulators.

#### CONCLUSION

MALS is a rare disease and it is easy to miss because it is not malignant, but patients can suffer from pain over a long period. For the accurate diagnosis of a patient complaining of abdominal pain, the diagnosis must be differentiated. In addition, as there are asymptomatic patients, patients who need treatment should be carefully selected, and improvement with medical treatment can be expected. Large-scale studies are also needed.

**Key Words:** Median arcuate ligamentum syndrome; Abdominal pain; Missed diagnosis; Neuromodulator; Celiac artery compression; Case report

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**Core Tip:** A considerable number of median arcuate ligamentum syndrome patients are overlooked in the diagnosis of patients complaining of abdominal pain. They are unable to lead a normal life due to severe excruciating pain. However, without an accurate diagnosis, they are misunderstood as symptomatic gallstone patients and have their gallbladder removed or experience pain for a long time. In addition, as the currently known treatment involves surgery based on a mechanistic hypothesis, there are concerns about its complications. Therefore, medical drug treatment should be prioritized to determine whether improvement occurs.

**Citation:** Kim JE, Rhee PL. Median arcuate ligamentum syndrome: Four case reports. *World J Clin Cases* 2022; 10(6): 1991-1997

**URL:** <https://www.wjgnet.com/2307-8960/full/v10/i6/1991.htm>

**DOI:** <https://dx.doi.org/10.12998/wjcc.v10.i6.1991>

## INTRODUCTION

Median arcuate ligamentum syndrome (MALS) is a disease entity with unclear pathogenesis[1,2]. If it is not considered in advance, the clinical diagnosis of the disease is very difficult because patients complain of digestive discomfort including pain, but this characteristic is not specific to MALS[3-6]. Most of the studies conducted through a retrospective analysis of incidental celiac artery compression in asymptomatic patients have been imaging studies[7-10], but the data are difficult to apply in actual clinical practice. This is because accidental image discovery does not require treatment. In addition, the surgical cases experienced by many surgeons only concern surgical methods for diseases that have already been diagnosed through complex processes[11,12], and no studies have been conducted to help make a quick diagnosis in these patients.

Therefore, it is necessary to introduce MALS as a causal disease, which should be considered in patients complaining of digestive symptoms at the gastroenterology department. Here, we present four patients diagnosed with MALS.

## CASE PRESENTATION

### Chief complaints

**Case 1:** A 63-year-old male patient with no specific medical history visited our clinic complaining of epigastric pain with heartburn symptoms that started 2 years earlier.

**Case 2:** A 64-year-old female patient, complaining of indigestion for 20 years had been treated for stress-induced gastritis, irritable bowel syndrome with constipation, and reflux esophagitis.

**Case 3:** A 62-year-old female patient with an underlying alcohol-related disorder complained of indigestion 3 d earlier and took digestive medicines, but there was no improvement.

**Case 4:** A 47-year-old female patient was admitted with severe abdominal pain that started several years ago.

### History of present illness

**Case 1:** The location of the pain was in the upper abdomen, and the pain lasted for 2 to 3 min. The patient mentioned that the pain intensity sometimes even woke him up and that it subsided when he walked and slightly improved when he bent over. Although he had been administered a proton pump inhibitor (PPI), motility-increasing agent, and mucosal protective agent at local clinics for several years, there was no improvement. Endoscopy and a computed tomography (CT) were already done at other hospitals. The CT readings at the other hospitals showed normal findings and malignancy was excluded.

**Case 2:** She underwent cholecystectomy at another hospital because of colicky pain, but she was still complaining of unbearable abdominal pain and came to our hospital. Endoscopy and a CT scan were

performed again and normal findings were reported.

**Case 3:** She visited the emergency room for severe abdominal pain but returned home without any specific findings on the CT scan. An outpatient clinic history was done again. Her squeezing pain persisted for more than 10 min at one time and was sometimes accompanied by chills.

**Case 4:** She felt excruciating pain in the upper abdomen that lasted several minutes to hours and the pain was not related to eating or defecation. Extreme pain even woke her up at night. It occurred once or twice a year, and PPI and prokinetics administration had no effect. The pain intensity decreased when leaning forward or breathing in.

### **History of past illness**

The patients had a free previous medical history.

### **Physical examination**

The patients' abdomen was soft and had no tenderness.

### **Laboratory examinations**

Blood analyses were normal.

### **Imaging examinations**

**Case 1:** We reviewed the medical history based on the characteristics of pain and the CT again, and a focal point of CT stenosis in the celiac axis was suspected (Figure 1A). Therefore, CT angiography (CTA) was performed. On the CTA, it was observed that the celiac trunk originating from the upper abdominal aorta was compressed by the diaphragmatic crus (Figure 1B). As a result, the origin of the celiac trunk showed focal tight narrowing, and along with clinical symptoms, the diagnosis could be made.

**Case 2:** Considering the patient's clinical features, we thought of diseases that could be missed by imaging techniques. Celiac artery compression was confirmed by reviewing the imaging studies (Figure 2A-C). This time, the collateral vessels around the pancreas were reviewed for narrowing of the celiac artery.

**Case 3:** CTA was performed based on clinical suspicion and celiac luminal narrowing was found due to focal calcification in the imaging tests (Figure 3A and B).

**Case 4:** There were no specific findings on endoscopy and abdominal ultrasonography, so based on clinical symptoms, an abdominal CT was performed, and MALS was diagnosed (Figure 4A-C).

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## **FINAL DIAGNOSIS**

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The final diagnosis of the presented four cases is median arcuate ligamentum syndrome.

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## **TREATMENT**

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### **Case 1**

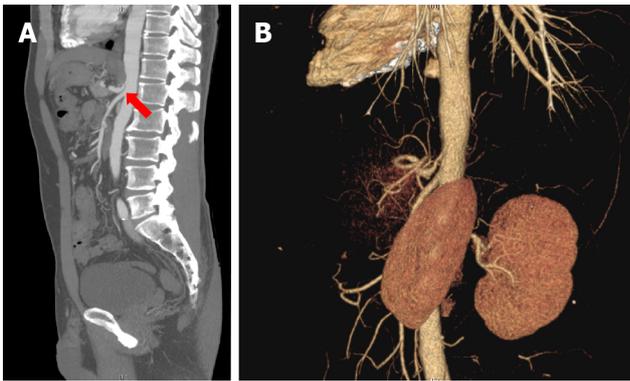
The patient's condition improved after the administration of amitriptyline 10 mg to treat visceral neurologic symptoms and Tegoprazan 50 mg. The patient showed an immediate symptom improvement response to amitriptyline before active analgesic administration. Thus, even before active treatment, through diagnosis alone, improvement was already demonstrated. During follow-up, if any pain remains after observation for more than 3 mo, a pain relief agent and an active agent for reducing neurotransmitters will be used.

### **Case 2**

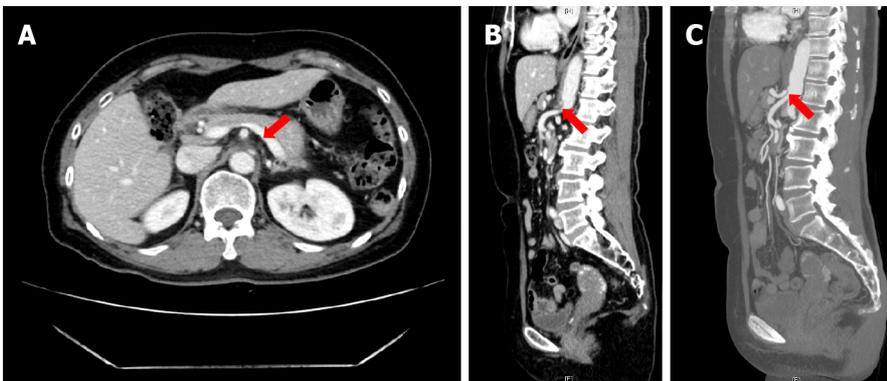
Neuropathic pain was relieved after the administration of gabapentin 100 mg, meloxicam 20 mg, and famotidine 20 mg b.i.d.

### **Case 3**

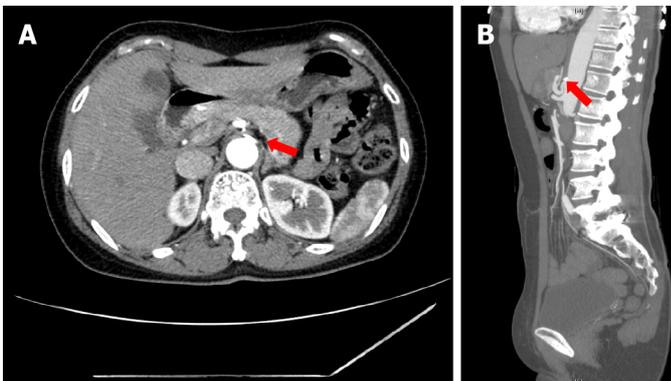
Gabapentin 100 mg, meloxicam 7.5 mg b.i.d, and esomeprazole were prescribed. She was due for an outpatient visit in 3 mo. The patient was asked to visit our hospital again if the symptoms worsened. She has been living without recurrence for a month after being prescribed medications.



**Figure 1** Imaging examinations of case 1. A: Computed tomography (CT) shows the celiac trunk originating from the upper abdominal aorta rather than in the normal case; B: CT angiography shows narrowing of the orifice of the celiac trunk due to compression by the diaphragmatic crus.



**Figure 2** Computed tomography shows the proximal celiac axis narrowing with collateral vessel formation. A: Axial view; B: Sagittal view; C: Computed tomography angiography confirmed the diagnosis.



**Figure 3** Computed tomography angiography shows mild luminal narrowing at the orifice of the celiac trunk with adjacent abdominal aortic calcification. A: Axial view; B: Sagittal view.

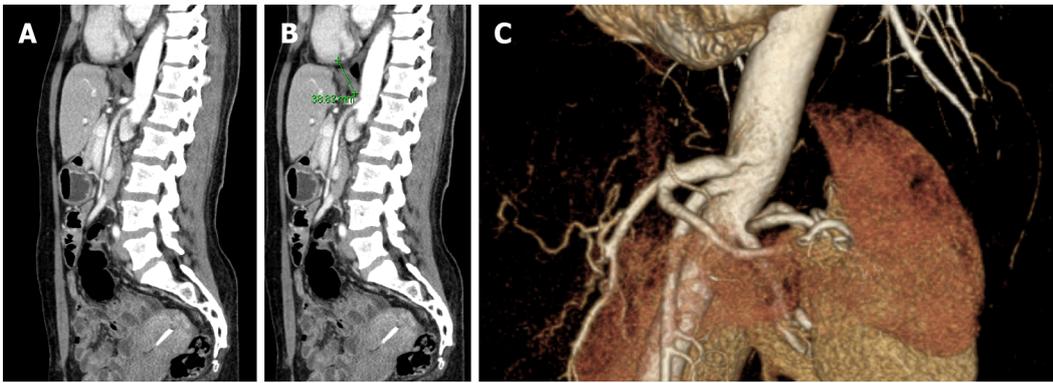
#### Case 4

She was prescribed gabapentin 100 mg, meloxicam 7.5 mg, and famotidine 20 mg b.i.d, and her symptoms improved significantly.

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### OUTCOME AND FOLLOW-UP

The symptoms of all 4 patients were much relieved with medical treatment, and they are being followed up on an outpatient clinics.



**Figure 4** Imaging examinations of case 4. A: Computed tomography (CT) showing the surrounding celiac trunk, where post-stenotic dilatation is seen; B: The median arcuate ligament compresses the narrowed area; C: CT angiography showing that the celiac trunk orifice is significantly narrowed, and post-stenotic dilatation is also seen.

## DISCUSSION

In all four cases, the patients experienced abdominal pain. In three cases, the diagnosis was possible by the judgment of the clinician shortly after the onset of symptoms. However, in one case that lasted over 20 years, a clear diagnosis was not possible. Even after complaining of colicky pain and performing a cholecystectomy, the diagnosis was made only after symptoms persisted. In all four cases, the symptoms were relieved by neuromodulators.

There are two current common hypotheses of MALS, which have been defined through several studies. The first one is neurologic pain caused by the compression of the celiac plexus and the other one is defined by compression of the celiac artery[2,10]. However, most hypotheses are based on images that were retrospectively analyzed and a defined protocol for making a diagnosis has not been developed. If this is due to ischemic arteriopathy, collateral vessels may develop, but the pain caused regardless of the produced collateral vessels. Importantly, ischemia cannot be reversible when the pain is maintained for a long time.

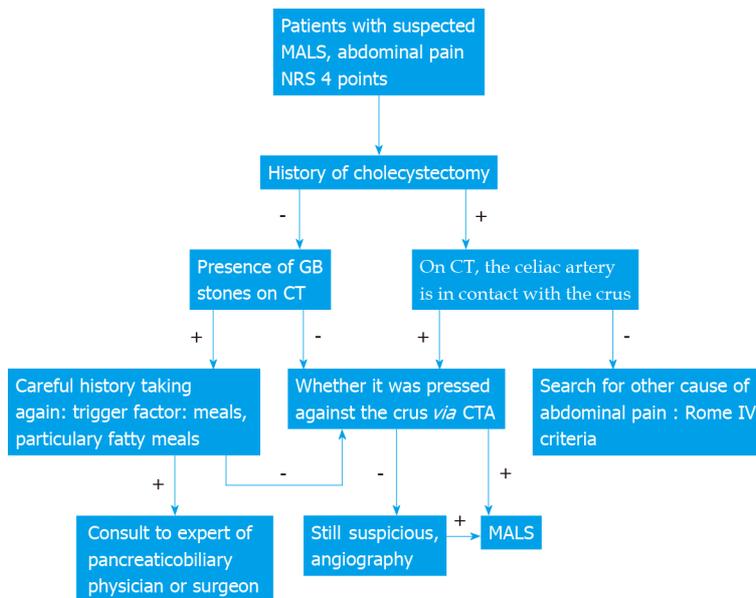
Therefore, the imaging diagnosis is only an auxiliary diagnosis. If the clinician does not suspect this diagnosis, the latter will happen very late. Most of the patients who came to the outpatient clinic of the department of gastroenterology at the tertiary hospital for indigestion and abdominal pain underwent an endoscopy and CT. However, it is not easy to identify rare diseases without providing clinical information from tens of thousands of still images. In addition, since the focus is on the exclusion of malignant tumors related to mortality pain is often considered a psychological disease despite its chronic nature, and is ignored.

A report in 2017 reported only one case[13]. Other case reports that dealt with diagnosed cases relied on images and explained the pathological mechanisms for bowel ischemia, but they could not explain the absence of symptoms of intestinal damage due to ischemia[14]. Surgical treatments, such as ganglionectomy along with arterial reconstruction, have been proven to be effective[11], but considering that there are no symptoms even when pressed, physical removal may not be the only solution. This case is very novel because no cases have reported the outcomes of medical treatment and it raises awareness about a diagnosis that should not be missed. This case was written to suggest that a gastroenterologist must make a differential diagnosis. And since the gastroenterologist judges all the clinical results together, the evaluation is performed after confirming these. The following algorithm illustrates this process (Figure 5).

A limitation of this study was that the number of cases studied was too small. However, given that numerous MALS diagnoses have been missed, it is necessary to review many patients in the future and establish the diagnostic flow, considering the possibility that the etiology itself may be wrong. Since considering only four cases is insignificant, it will be necessary to review patients experiencing abdominal pain who have numerous missed diagnoses in the future. A common diagnostic flow is also required.

## CONCLUSION

MALS is a rare disease and it is easy to miss because it is not malignant, but patients experience pain over a long period. For the accurate diagnosis of a patient complaining of abdominal pain, the diagnosis must be differentiated. In addition, as there are asymptomatic patients, patients who need treatment should be carefully selected, and improvement with medical treatment can be expected. Large-scale studies are also needed.



**Figure 5 Algorithm for median arcuate ligamentum syndrome diagnosis through imaging modalities and clinical history.** MALS: Median arcuate ligamentum syndrome; NRS: Numeric pain rating scale; GB: Gallbladder; CT: Computed tomography; CTA: Computed tomography angiography.

## FOOTNOTES

**Author contributions:** Rhee PL did study concept and design; Kim JE did data acquisition, drafting of the manuscript and critical revision of the manuscript for important intellectual content; all authors did approval of final manuscript.

**Informed consent statement:** All case participants provided informed written consent for publish.

**Conflict-of-interest statement:** No potential conflict of interest relevant to this article was reported.

**CARE Checklist (2016) statement:** The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

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**S-Editor:** Zhang H

**L-Editor:** A

**P-Editor:** Zhang H

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