

## CASE REPORT ΕΝΔΙΑΦΕΡΟΥΣΑ ΠΕΡΙΠΤΩΣΗ

# Median arcuate ligament syndrome in a 41-year-old woman

Median arcuate ligament syndrome, also called celiac artery compression syndrome or Dunbar syndrome, is an uncommon cause of post-prandial abdominal pain and aneurysms of the pancreaticoduodenal arcade. The nonspecific manifestations of the syndrome hinder clinical suspicion and make the diagnosis a challenging condition of exclusion. Diverse resources of abdominal imaging evaluations have been utilized to establish the correct diagnosis, and the options for surgical management aim to perform the median arcuate ligament release and the transection of the celiac plexus as the effective solution.

Median arcuate ligament syndrome (MALS), also known as Dunbar syndrome, involves narrowing of the celiac artery root by MAL compression, pancreaticoduodenal artery aneurysm (PDAA), and postprandial pain. The incidence estimated is two cases per 100,000 population.<sup>1-10</sup> Diagnosis of MALS is often one of exclusion, and the open, laparoscopic, or retroperitoneoscopic surgeries have been the treatment option with good outcomes.<sup>1-10</sup> These definitive surgical procedures should include the complete division of the MAL fibers after reaching the root of the celiac artery

from the pancreatic superior border.<sup>7</sup> Confirmed diagnosis and surgical management of MALS is described in an adult female. The aim is to enhance the suspicion index among the non-specialist healthcare workers.

### CASE PRESENTATION

A 41-year-old woman with a history of kidney stones, hysterectomy, and tonsillectomy sought medical care complaining of a feeling of nausea for about two years, "as if she had eaten too

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L.A.M. dos Santos,<sup>1</sup>  
V.M. dos Santos,<sup>2</sup>  
P.S. Scomparin,<sup>3</sup>  
J.C.V. Rios,<sup>4</sup>  
A.P. Metzger,<sup>5</sup>  
V.R.D.A. Modesto<sup>6</sup>

<sup>1</sup>General Surgery and Advanced Program of Oncosurgery and Videolaparoscopy, Public Workers State Hospital Francisco Morato de Oliveira, São Paulo-SP

<sup>2</sup>Department of Internal Medicine, Armed Forces Hospital and Catholic University, Brasília-D

<sup>3</sup>Digestive Surgery, Integrated Medical Clinic, University of Marília, Marília-SP

<sup>4</sup>Digestive Surgery, Santa Paula Hospital, São Paulo-SP

<sup>5</sup>Medical Course, University Center of the Americas, São Paulo-SP

<sup>6</sup>Medical Course, Faculty of Medical Sciences, Holy House of São Paulo, São Paulo-SP, Brazil

Σύνδρομο μέσου τοξοειδούς  
συνδέσμου σε γυναίκα 41 ετών

Περίληψη στο τέλος του άρθρου

### Key words

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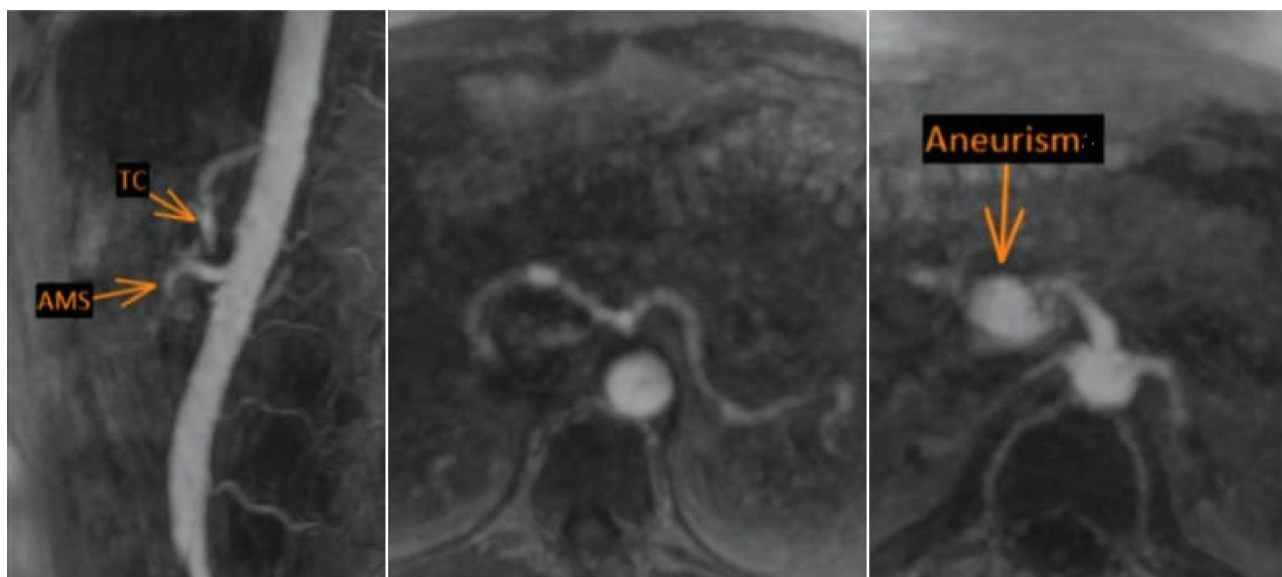
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much and her stomach felt stiff". She denied vomiting episodes. After breakfast, she reported a feeling of "heaviness in her stomach" throughout the day, which worsened after eating any kind of food. She emphasized the severe abdominal discomfort, manifested by a constant feeling of distension; besides, she denied vomiting, had episodes of diarrhea, but no weight loss, and was unaware of another similar case in her family.

The physical examination revealed that she was in good general condition, oriented, flushed, hydrated, afebrile, anicteric. Breath sounds were normal with no adventitious sounds. The abdomen was soft and painless on palpation, without visceromegaly or palpable masses, peritoneal irritation and abnormal bowel sounds. Her body weight was 67 kg, her height: 1.60 m, body mass index (BMI): 26.17 kg/m<sup>2</sup>, heart rate 86 bpm, respiratory rate 14 ipm, arterial blood pressure 116/82 mmHg, O<sub>2</sub> saturation in ambient air 98%, and axillary temperature 36.5 °C. Laboratory determinations showed red cells count: 4.29 million/ $\mu$ L, hemoglobin: 12.1 g/dL, hematocrit: 36.3%, MCV: 84.6 fL, MCH: 28.2 pg, MCHC: 33.3 g/dL, RDW: 13.3%, leukocytes: 13,680/ $\mu$ L, segmented: 13,133/ $\mu$ L, lymphocytes: 410/ $\mu$ L, monocytes: 137/ $\mu$ L, platelets: 289,000/ $\mu$ L, urea: 33 mg/dL, creatinine: 0.98 mg/dL, sodium: 136 mmol/L, potassium: 4.7 mmol/L, and the glomerular filtration rate (GFR): 75 mL/min/1.73m<sup>2</sup>. Abdominal computed tomography (CT) and angiography showed an accentuated stenosis of the celiac trunk due to compression of the arcuate ligament of the diaphragm, in addition to saccular aneurysms from the inferior pancreaticoduodenal artery (fig. 1). Additionally, the abdominal and pelvic ultrasonography (US) evaluation revealed normal liver, gallbladder, pancreas, spleen, and kidneys, besides the bilateral renal lithiasis. Arterial magnetic resonance angiography showed the aorta and celiac trunk presenting with normal aspects, compression by

MAL, and the inferior PDAA. Under routine conditions of general anesthesia, the patient underwent a supraumbilical incision with release of the umbilical scar, and creation of pneumoperitoneum with a Veres needle and passage of an 8-mm catheter and Da Vinci  $\times$  DE 30 camera, in addition to 8 mm trocars (one left lateral and two right lateral) under direct vision. The following forceps were used: Cadiere forceps (fenestrated), bipolar fenestrated, and permanent cautery hook from right to left.

Access was gained via the lesser gastric curvature, with opening of the gastro-hepatic ligament using the monopolar hook forceps, and opening by layers after separation with the Cadiere forceps, and robot-assisted lysis of the blood. There was identification of the left gastric artery with isolation, identification of the splenic artery and celiac trunk, and identification of the supra-celiac aorta with fiber release. Color Doppler was utilized to identify the visceral arteries, distinguishing the MAL; the ligament fibers were sectioned using the monopolar hook energy until complete release at the emergence of the celiac trunk. The abdominal wall was reconstructed by a myocutaneous flap, clamps and trocars were removed, and pneumoperitoneum was released. Umbilical sutures were performed by Vicryl 0, while portal site closures were performed by Monocryl 4.0. The anatomopathological conclusive diagnosis was MALS, and was based on the evaluation of two fragments of tissue measuring 1.5 $\times$ 0.6 $\times$ 0.3 cm, with lobulated external surfaces and presenting with a yellowish-white color and a firm and elastic consistency. Notably, the histopathological study revealed adipose tissue containing nerve fibers and foci of fibrosis, with no evidence of malignancy or cell atypia. Physical examination after the robotic surgery revealed the good-looking scars, and her restricted complaints were mild local discomfort compatible with the healing process. Postoperative follow-up was



**Figure 1.** Arterial magnetic resonance angiography of the upper abdomen showing normal aorta, celiac trunk (TC) with subocclusion due to extrinsic compression by the median arcuate ligament, and filling after approximately 1.1 cm through collaterals; saccular aneurysm (approximately 2.5 cm) in the inferior pancreaticoduodenal artery.

unremarkable, and abdominal and pelvic CT for control revealed some densification around the celiac trunk associated with the surgical procedure; liver, biliary tract, pancreas, and spleen without alterations, absence of obstructive uropathy, and the presence of multiple bilateral kidney stones. At her hospital discharge, she was recommended to maintain a longstanding follow-up.

## COMMENTS

Arino et al described a 54-year-old man with epigastric pain, vomiting, and difficulty maintaining oral intake, symptoms similar to his previous admission four years earlier.<sup>1</sup> At that time, abdominal CT revealed small aneurysms in branches of the pancreaticoduodenal artery communicating with the first jejunal artery, and stenosis at the origin of the celiac artery, besides aneurysm rupture and hematoma related to MALS. Recent images confirmed the PDAA rupture, and the prompt arterial coil embolization was done; he was discharged home with normal ingestion on the third postoperative day. He remained on the outpatient follow-up with abdominal imaging, over 22 months. The authors highlighted the role of the long-term follow-up of PDAA related to MALS due to the possibility of some future rupture or re-rupture of the associated aneurysms.<sup>1</sup>

Dung et al reported a 66-year-old female presenting postprandial epigastric pain for four years, not relieved by treatment of the gastritis diagnosed by endoscopic evaluation.<sup>2</sup> One year later, the abdominal CT revealed hook-shaped stenosis of the celiac trunk and mild post-stenosis dilatation, classically indicative of the diagnosis of MALS; the patient refused the proposed surgery, and was only submitted the angioplasty with a stenting.<sup>2</sup> During the five months of follow-up, there was no gastrointestinal disturbance; the authors stressed angioplasty and stenting for MALS with a celiac trunk atherosclerosis.<sup>2</sup>

Hu et al described a 77-year-old man with epigastric pain during five months, with abdominal ultrasound and gastroenterological endoscopy examinations being unremarkable.<sup>3</sup> A celiac artery angiography showed 90% narrowing, and revascularization was done; abdominal pain recurred six months later, and CT angiography showed the obstructed stent. As the patient did not accept decompression of the celiac artery, the post-dilatation was performed, with less than 50% of residual stenosis and a resolution of abdominal pain.<sup>3</sup>

Hussen et al reported a 29-year-old woman with postprandial abdominal pain for five months; and CT angiography scan and Doppler US showed MALS and cholelithiasis.<sup>4</sup> Laparoscopy procedure confirmed the compression of the

aorta and celiac trunk near its origin; the ligament was opened up by a scalpel, and the cholecystectomy was done with success. In the three-month follow-up, she had no postprandial abdominal symptoms.<sup>4</sup>

Kiudelis et al described a 40-year-old woman with chronic postprandial abdominal pain and weight loss; the CT angiography showed the celiac artery with 50% stenosis.<sup>5</sup> Laparoscopic release of the MAL was performed by a harmonic dissector, with no postoperative complications or symptoms, and hospital discharge was on the third day. With better appetite and weight gain, her CT of control showed no artery compression.<sup>5</sup>

Manchella et al reported a case of a 51-year-old woman with numerous visceral aneurysms involving the pancreaticoduodenal arcade associated with MALS-related compression.<sup>6</sup> She had chronic postprandial abdominal pain and evidence of cholelithiasis (treated by cholecystectomy), but symptoms recurred shortly thereafter, and MALS was diagnosed. During the laparotomy the celiac artery was found compressed by diaphragmatic fibers, which were sectioned to obtain a complete MAL release of the vascular compressions. The common hepatic artery was then isolated and skeletonized to facilitate anastomosis.<sup>6</sup> Six days after revascularization, endovascular embolization was performed, and a control CT angiogram performed one month postoperatively showed a patent aorto-celiac bypass graft and occlusion of the large aneurysms arising from the superior mesenteric artery.<sup>6</sup>

Nishino et al described a 67-year-old male with an antecedent of distal gastrectomy and D2 lymph node dissection for gastric cancer, and 16 years later had an acute abdomen due to cholangitis and pancreatitis, besides the MALS with retroperitoneal hematoma.<sup>7</sup> Percutaneous transhepatic cholangial drainage was performed with hematoma shrinkage and improvement in cholangitis and pancreatitis. The patient was then referred for surgical intervention, through which the root of the celiac artery from the inferior border of the pancreas was accessed, successfully dividing the MAL, and confirming the improved blood flow.<sup>7</sup>

Tanikawa et al reported a 56-year-old woman with acute abdomen by rupture of a pseudoaneurysm, associated with MALS. Abdominal CT examinations showed a retroperitoneal hematoma, celiac artery stenosis, and two aneurysms of inferior PDA.<sup>8</sup> Hemostasis was performed by transcatheter arterial embolization, but 15 days later, there was duodenal stenosis with gastric distension and nausea; as the conservative approach was not effective, the patient underwent the MAL transection and gastrojejunostomy.<sup>8</sup> Her four-month follow-up showed complete resolution of

duodenal stenosis. The authors emphasized the role of early diagnosis and a multidisciplinary management of MALS.<sup>8</sup>

Wang et al compared data from 27 MALS patients and 427 age- and sex-matched controls, 69.1% male with a median age of 59 years, from January 2021 to July 2023.<sup>9</sup> An age- and sex-matched hypertension cohort was a control group for comparison, classified into mild-to-moderate and severe stenosis based on a 50% stenosis threshold.<sup>9</sup> Imaging studies demonstrated increased abdominal aorta and distal superior mesenteric artery (SMA) diameters in the MALS group; higher age, alcohol consumption, lower triglycerides level, and larger SMA diameter were associated with the MALS severity.<sup>9</sup> The authors concluded that the incidence of severe complications among MALS patients does not correlate significantly with age and peaks in those from 60–69 years (50.8%).<sup>9</sup>

Yadav et al reported a case of a 51-year-old man presenting a postprandial abdominal pain for ten years, and a CT angiogram showing celiac artery changes consistent with MALS.<sup>10</sup> He underwent laparoscopic division of the MAL, decompressing the celiac trunk, and the symptoms

did not recur with the normal oral intake; discharge was one week later, and follow-up controls until three months after the procedure revealed no abnormalities.<sup>10</sup> The authors highlighted the challenging diagnosis of MALS due to nonspecific symptoms and the major role of celiac trunk compression images to confirm this rare condition, whose definitive treatment requires the prompt and complete surgical decompression.<sup>10</sup>

In conclusion, MALS is an uncommon and yet incompletely understood potentially severe condition. Due to the low prevalence and nonspecific manifestations, healthcare professionals should have a high index of suspicion for MALS in cases of postprandial abdominal pain. One must consider MALS after exclusion of intestinal disorders by imaging evaluations. Early detection and prompt surgical intervention increase favorable outcomes for patients with MALS, with decompression of the celiac artery constituting the cornerstone of treatment; therefore, ligament release combined with transection of the celiac plexus is considered an effective management. Long-term follow-up is essential for secondary PDAA due to later recurrence and rupture.

## ΠΕΡΙΛΗΨΗ

### Σύνδρομο μέσου τοξοειδούς συνδέσμου σε γυναίκα 41 ετών

L.A.M. DOS SANTOS,<sup>1</sup> V.M. DOS SANTOS,<sup>2</sup> P.S. SCOMPARIN,<sup>3</sup> J.C.V. RIOS,<sup>4</sup> A.P. METZGER,<sup>5</sup> V.R.D.A. MODESTO<sup>6</sup>

<sup>1</sup>General Surgery and Advanced Program of Oncosurgery and Videolaparoscopy, Public Workers State Hospital Francisco Morato de Oliveira, São Paulo-SP, <sup>2</sup>Department of Internal Medicine, Armed Forces Hospital and Catholic University, Brasília-DF, <sup>3</sup>Digestive Surgery, Integrated Medical Clinic, University of Marília, Marília-SP, <sup>4</sup>Digestive Surgery, Santa Paula Hospital, São Paulo-SP, <sup>5</sup>Medical Course, University Center of the Americas, São Paulo-SP, <sup>6</sup>Medical Course, Faculty of Medical Sciences, Holy House of São Paulo, São Paulo-SP, Βραζιλία

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Το σύνδρομο συμπίεσης του μέσου τοξοειδούς συνδέσμου, που ονομάζεται επίσης σύνδρομο συμπίεσης της κοιλιακής αρτηρίας ή σύνδρομο Dunbar, είναι μια ασυνήθιστη αιτία μεταγευματικού κοιλιακού άλγους και ανευρυσμάτων του παγκρεατοδωδεκαδακτυλικού τόξου. Οι μη ειδικές εκδηλώσεις του συνδρόμου δυσχεραίνουν την κλινική υποψία και καθιστούν τη διάγνωση μια δύσκολη προϋπόθεση αποκλεισμού. Έχουν χρησιμοποιηθεί ποικίλες πηγές αξιολογήσεων κοιλιακής απεικόνισης για την τεκμηρίωση της σωστής διάγνωσης και οι επιλογές για χειρουργική αντιμετώπιση στοχεύουν στην εκτέλεση της απελευθέρωσης του μέσου τοξοειδούς συνδέσμου και της διατομής του κοιλιακού πλέγματος ως αποτελεσματική λύση.

**Λέξεις ευρητηρίου:** Σύνδρομο μέσου τοξοειδούς συνδέσμου, Σύνδρομο Dunbar, Σύνδρομο συμπίεσης κοιλιακής αρτηρίας

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*Corresponding author:*

V.M. dos Santos, Armed Forces Hospital, Estrada do Contorno do Bosque s/n, Cruzeiro Novo, CEP 70630-900, Brasília-DF, Brazil  
e-mail: vitorinomodesto@gmail.com