Case report
Prikaz bolesnice

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LAPAROSCOPIC TREATMENT OF MEDIAN ARCUATE LIGAMENT SYNDROME – A CASE REPORT

Laparoskopsko liječenje sindroma medijalnog lučnog ligamenta – prikaz bolesnice

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Abstract

This report describes the case of a 42-year-old woman who suffered from postprandial abdominal pain, bouts of tachycardia, belching, tinnitus, and syncope interfering with everyday life.
After an extensive diagnostic workup, the correct diagnosis was finally established by computed tomography angiography. The patient had a syndrome caused by compression of the celiac trunk by the median arcuate ligament, also known as Dunbar syndrome. It typically affects younger patients, particularly women, with the triad of postprandial abdominal pain, weight loss, and abdominal bruit. The patient was successfully treated with laparoscopic decompression of the celiac trunk and resection of the celiac plexus. The median arcuate ligament syndrome should be included in the differential diagnosis for young patients presenting with postprandial epigastric pain, food fear, unintentional weight loss, and normal upper gastrointestinal endoscopy findings.

Sažetak

U ovom radu prikazujemo slučaj 42-godišnje bolesnice s bolovima u trbuhu nakon jela, epizodama tahikardije, podrigivanja, tinitusa i sinkopa, što je značajno narušavalo kvalitetu života i svakodnevno funkcioniranje.

Ključne riječi:
sindrom medijalnog lučnog ligamenta; sindrom kompresije celijačnog debla; Dunbarov sindrom; laparoskopija

Nakon opsežnih dijagnostičkih pretraga koje su bile uredne, točna dijagnoza konačno je postavljena CT angiografijom. Utvrđen je sindrom kompresije celijačnog debla medijalnim lučnim ligamentom (MALS - engl. median arcuate ligament syndrome), poznat i kao Dunbarov sindrom. Ovaj sindrom najčešće pogađa mlađe bolesnike, osobito žene, a karakterizira ga trijas bolova u trbuhu nakon obroka, gubitak tjelesne mase i čujni šum kod auskultacije gornjeg dijela trbuha.
Bolesnica je uspješno liječena laparoskopskom dekompresijom celijačnog debla, uz resekciju celijačnog pleksusa. Sindrom medijalnog lučnog ligamenta treba razmotriti u diferencijalnoj dijagnostici mlađih bolesnika s postprandijalnom epigastričnom boli, strahom od uzimanja hrane, nenamjernim gubitkom tjelesne mase i urednim nalazom gastroskopije.

INTRODUCTION

The median arcuate ligament syndrome (MALS), also known as Dunbar syndrome, is a vascular compression syndrome caused by compression of the celiac trunk by the median arcuate ligament (MAL) due to an anatomic variation in the origin of the artery. The syndrome triggers the triad of postprandial abdominal pain, weight loss, and abdominal bruit. Furthermore, compression and narrowing of the celiac trunk lumen may result in symptomatic ischemia of the involved organs.[1,2]

MALS is one of several rare syndromes in abdominal surgery that are notoriously difficult to diagnose but could have a significant effect on the quality of life; therefore, it deserves wider attention. It often affects young patients, especially females. The diagnosis is elusive, considering that the nonspecific symptoms overlap with other gastrointestinal pathologies. Therefore, patients with MALS usually undergo extensive and repetitive workups, including psychological and psychiatric assessments, before reaching the correct diagnosis.

Herein, we present the case of a 42-year-old female patient with an extensive medical workup, but only computed tomography angiography (CTA) provided the key to the correct diagnosis.

This case report has been reported in line with the SCARE Criteria.[3]

CASE DESCRIPTION

A 42-year-old woman had experienced postprandial abdominal pain since giving birth eight years earlier. Besides the epigastric pain after meals, she suffered from sudden bouts of tachycardia, tinnitus, belching, weakness, and syncope, significantly impairing the patient's quality of life. The patient complained of frequent yawning, air hunger, and belching while talking. Gradually, she limited her food intake to lessen the symptoms. At the moment of diagnosis, the patient was taking diazepam only, although she had previously tried various medications. The patient was an active smoker and had a history of laparoscopic bilateral salpingectomy.

An extensive medical examination was undertaken: a complete upper and lower gastrointestinal endoscopy, neurological and psychological evaluation, radiology imaging, including abdominal ultrasound and lung computed tomography (CT) as a part of an immunology workup. As liver lesions were seen on a lung scan, the clinician decided to proceed with abdominal CTA, which shed light on the correct diagnosis. The lesions in the liver were hemangiomas.

Diagnostic Assessment

The multiphase contrast CT scan demonstrated a low insertion of the MAL, causing significant narrowing of the celiac trunk and post-stenotic trunk

dilatation more distally (Figure 1). Doppler ultrasound of the celiac trunk, a dynamic imaging tool performed before surgery in forced inspiration and expiration, corroborated the diagnosis of MALS. At rest, peak systolic velocity in the celiac trunk reached 387 cm/s. During inspiration, the velocity was 272 cm/s, whereas the expiratory exam recorded a velocity greater than 532 cm/s. This objective finding confirmed dynamic compression of the celiac trunk depending on the MAL movement during respiration. Complete laboratory findings were within the reference values. A physical examination revealed a typical bruit over the epigastric region, which was noted only after the diagnosis was reached. Her BMI was 22.1 kg/m^2, weight 61 kg, height 166 cm.

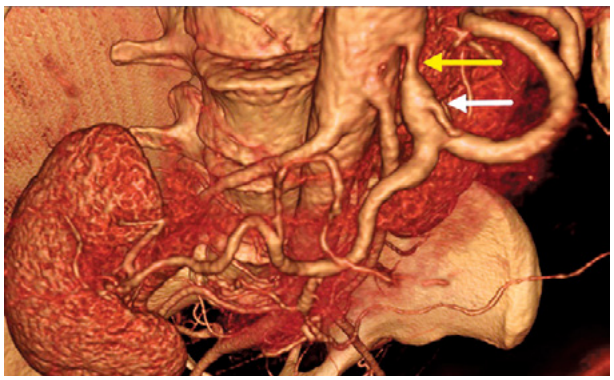


Figure 1 Three-dimensional reconstruction of computed tomography angiography (CTA) demonstrating significant celiac trunk stenosis (yellow arrow) and post-stenotic dilatation (white arrow)
Slika 1. *Trodimenzionalna rekonstrukcija CT angiografije s vidljivom značajnom ishodišnom stenozom celijačnog debla (žuta strelica) i poststenotičkom dilatacijom (bijela strelica)*

Intervention and follow-up

The patient was scheduled for laparoscopic decompression of the celiac trunk. After routine preoperative assessment, the patient underwent an uneventful laparoscopic decompression of the celiac trunk and MAL resection under general anesthesia. Pneumoperitoneum was created with a Veress needle inserted through Palmer's point. The 30-degree endoscope was inserted through a 10 mm umbilical port. An additional 10 mm and three 5 mm trocars were inserted bilaterally subcostally, as shown in Figure 2. The pars flaccida of the lesser

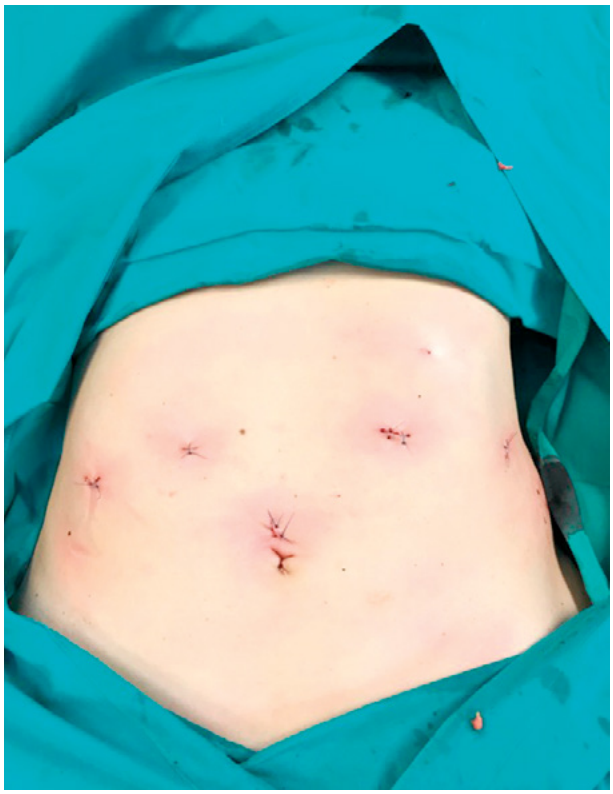


Figure 2 Postoperative image of the abdominal wall demonstrating a minimally invasive approach and the position of five laparoscopic ports
Slika 2. *Poslijeoperacijska fotografija trbušne stijenke koja prikazuje minimalno invazivni pristup i poziciju laparoskopskih troakara*

omentum was incised first, enabling entry to the omental bursa. After the root of the left gastric artery was isolated, the fibers of the diaphragmatic crura anterior to the aorta were resected until the exposure of the aortic adventitia. Resection of all

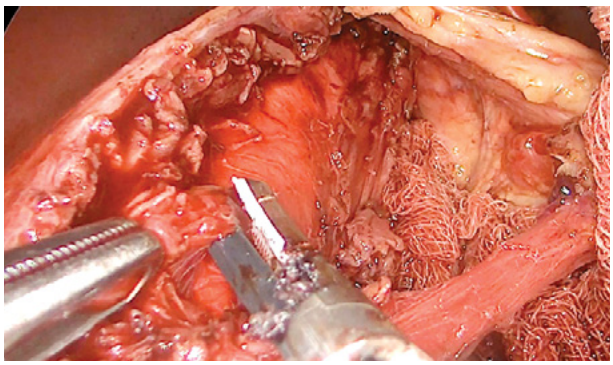
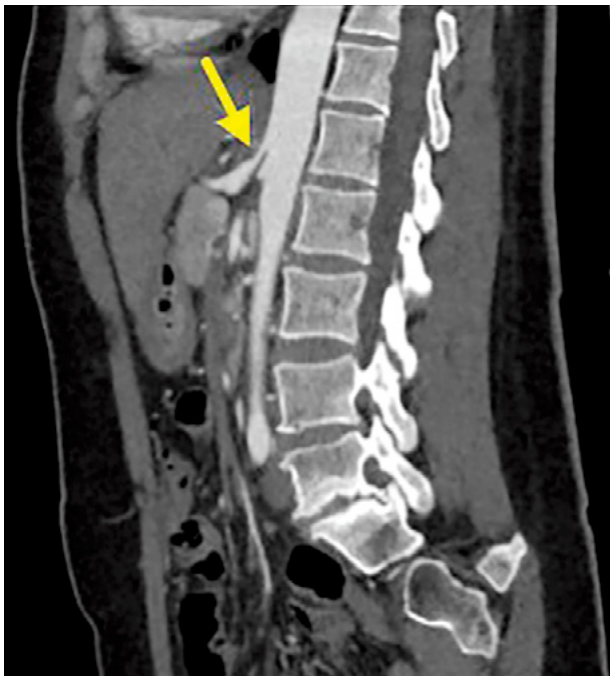


Figure 3 Intraoperative view of the harmonic scalpel transecting the fibers of the median arcuate ligament over the root of the celiac trunk
Slika 3. *Intraoperacijska fotografija harmoničnog skalpela kojim se režu vlakna medijalnog lučnog ligamenta iznad ishodišta celijačnog debla*

fibers caudally was carefully continued, reaching the root of the celiac trunk being severely stenosed under the fibers of the median arcuate ligament, resembling tightened strings. The celiac trunk was freed entirely (Figure 3). Furthermore, fibers of the celiac plexus crossing the origin of the celiac trunk and the aorta were divided. The diaphragmatic crura were approximated with two non-resorbable sutures, thereby calibrating the hiatus appropriately to prevent hiatal herniation.



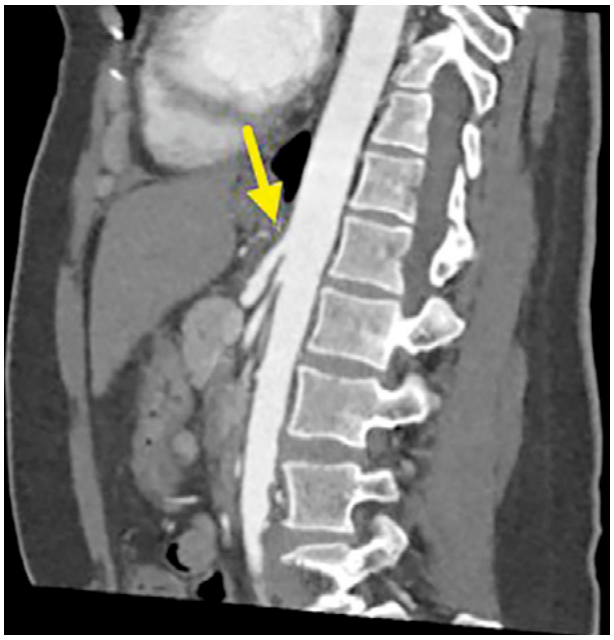


Figure 4 (A): Preoperative CTA, sagittal view, arrow pointing to stenosis of the celiac trunk. (B): Postoperative CTA, sagittal view, arrow pointing to the normal caliber of the celiac trunk root
Slika 4. *(A): Prijeoperacijska CT angiografija, sagitalni prikaz – strelica pokazuje stenozu celijačnog debla. (B): Poslijeoperacijska CT angiografija, sagitalni prikaz – strelica pokazuje uredan promjer ishodišta celijačnog debla*

Postoperative recovery was uneventful, with rapid restoration of peristalsis, tolerating early oral intake. Postoperatively, a Doppler ultrasound of the celiac trunk showed improved celiac blood flow with diminished influence of diaphragmatic excursions during respiration. At rest, peak systolic velocity was 229 cm/s, during expiration 264 cm/s, and 241 cm/s in inspiration. These values approached the established reference ranges.[4-6] The patient was discharged on postoperative day 3. Follow-up CTA one year after surgery confirmed a pristine patency of the celiac trunk without previously visible stenosis (Figure 4). The patient gained two kilograms after surgery. Her symptoms were significantly reduced, and yawning and belching stopped.

DISCUSSION

In 1917, Lipshutz first described celiac trunk compression.[7] In 1963, Harjola described MALS, while the first MALS clinical study was published by Dunbar et al. two years later.[8-9] Since then, numerous clinical studies and case reports have been published. MALS is a rare condition with uncertain true epidemiology, with estimate occurrence of two cases per 100,000 persons. It commonly affects young patients, especially females.[10] The case presented herein confirms the rule that MALS is a diagnosis of exclusion. Considering that, after extensive workup, only CTA provided the key to the correct diagnosis.

Since the first case of successful surgery was published in the 1960s, an open approach has been largely abandoned thanks to the emergence of minimally invasive surgery.[11] In the era of open surgery, the diagnosis of MALS and the decision to perform surgical treatment were challenging due to limited imaging techniques, including the lack of the CTA, as well as the wide range of po-

tential postoperative complications and significant morbidity.[12] Minimally invasive access, such as laparoscopic or robotic surgery allow enhanced visualization of the anatomy, reduced postoperative pain, and fast recovery.[13,14] The length of stay is short, and morbidity is minimal, allowing both the surgeon and patient to make a more informed decision about the surgical treatment. When a judiciously selected patient with a convincing CTA and corresponding symptoms undergoes surgical decompression, immediate relief is achieved. It should be emphasized that MALS is a rare condition, and the technical feasibility of decompression alone does not justify the indications should be overly liberal and inappropriate. A comprehensive diagnostic workup is mandatory to avoid missing a more common pathology. Published cases describe the confirmed MALS diagnosis of a patient later diagnosed with Crohn's disease.[15,16] Moreover, other authors have described cases of MALS in patients with underlying ulcerative colitis or pancreatic cancer.[17,18] Importantly, unrecognized MALS can cause serious postoperative complications in major abdominal surgeries, such as liver transplantation or pancreaticoduodenectomy, due to ischemia of the liver and stomach, highlighting the need for a complete preoperative vascular assessment.[19-22] The celiac trunk compression syndrome may be suspected based on the triad of postprandial abdominal pain, weight loss, and abdominal bruit. Nevertheless, finding the correct diagnosis can be daunting and challenging; therefore, early vascular imaging is recommended. It is crucial that, once MALS is suspected, patients undergo relevant imaging, primarily Doppler ultrasound, which is a cost-effective, sensitive imaging modality that does not involve ionizing radiation. Positive Doppler for MALS should be confirmed with abdominal CTA, which is necessary in planning surgical decompression.

Clinicians and researchers should be aware of the symptoms that may suggest MALS, especially since it is a rare diagnosis of exclusion, often confirmed only after patients undergo comprehensive but exhausting testing.

Informed consent

Written informed consent was obtained from the patient for the publication of this case report and accompanying images.

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