



## Case report

Laparoscopic treatment of symptomatic Dunbar syndrome: a case report<sup>☆</sup>

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## ABSTRACT

**Introduction and aim of the study:** Dunbar syndrome is a rare disorder due to the external compression of the celiac trunk by the median arcuate ligament. The symptoms include abdominal pain, nausea and vomiting with a significant reduction in patients' quality of life. Laparoscopy has proven to be a safe procedure. The aim of this study is to present a case of Dunbar syndrome undergoing laparoscopic surgery.

**Case presentation:** A 40-year-old male patient presented at Emergency Room with upper abdominal pain and dyspepsia, related to food intake. A selective arteriography of the celiac trunk revealed stenosis due to compression of the celiac artery.

**Clinical discussion:** Laparoscopic decompression of the celiac trunk by the median arcuate ligament was performed. Postoperative course was uneventful and the patient was discharged on the 5th postoperative day.

**Conclusion:** Minimally invasive surgical division of the arcuate ligament is feasible and safe and it can restore patients' quality of life.

## 1. Introduction

Dunbar syndrome, also known as celiac artery compression syndrome or median arcuate ligament syndrome (MALS) is a rare disease, characterized by a pathological insertion of the median arcuate ligament at a lower level of the diaphragm, narrowing the lumen of the celiac trunk by compression [2].

Dunbar syndrome is more prevalent in children and adolescents and is associated with vegetative symptoms, mainly during expiration. It was first described by Harjola in 1963 and Dunbar et al. in 1965: the incidence of MALS is unknown, but in the last years case reports have increased, possibly due to the widespread use of multiplanar contrast enhanced imaging [3].

The symptoms include the classical triad observed in mesenteric ischemia, characterized by postprandial abdominal pain, nausea and vomiting and subsequently weight loss [4,5]. The heterogeneity of symptoms explains why the diagnostic process is not so clear and patients often are addressed to several specialists before consulting to the

surgeon. After the suspicion, selective angiography in expiration or magnetic resonance angiography can identify the stenosis of the initial segment of the celiac artery and confirm the diagnosis. The treatment includes the section of the median arcuate ligament and the fibers of the celiac plexus that can be performed by percutaneous transluminal angioplasty or surgery [4,5]. Since laparotomic surgery is considered an invasive technique, laparoscopy has been proposed as an alternative approach [6,7]. The aim of this report is to present a case of Dunbar syndrome treated by laparoscopic approach. The work was written in line with the SCARE criteria [1]. Consent to the processing of data for scientific purposes is requested and signed at the time of admission and kept in the medical record.

## 2. Case presentation

In March 2021 a 40-year-old male presented to Emergency Department with one-month history of post-prandial diffuse abdominal pain and dyspepsia. His past medical history included laparotomic

**Abbreviations:** MALS, median arcuate ligament syndrome; SMA, superior mesenteric arteria; USG, ultrasonography; CECT, contrast enhanced computed tomography; PSV, peak systolic velocity; IA, invasive angiography.

<sup>\*</sup> The work was written in line with the SCARE criteria (Agha et al., 2020) [1]. Consent to the processing of data for scientific purposes is requested and signed at the time of admission and kept in the medical record.

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appendectomy and fibromyalgia in rheumatologic follow-up. Blood tests were normal, without LDH increase. A contrast enhanced computed tomography (CECT) of the abdomen was performed, revealing a shrinkage of SMA origin without pneumoperitoneum or liquid layer (Fig. 1).

He was then admitted to the Surgical Department and treated with liquid only diet and pain-killers. He underwent an elective USG Doppler, that confirmed a focal significant narrowing at the origin of SMA with a peak systolic velocity (PSV). Invasive angiography (IA) was performed showing a moderate stenosis of the SMA origin together with a severe obstruction of the celiac trunk origin. A retrograde compensation of the blood flow was also reported (Fig. 2).

An interventional radiology consult was then asked for possible stenting of the celiac artery, which was considered not indicated in this case since symptoms were attributed mainly to SMA compression. In view of his long-lasting symptoms, causing significant food intake restrictions, surgical laparoscopic treatment of the condition was proposed to the patient (Fig. 3).

The surgery consisted in the section of the median arcuate ligament and the fibers of the celiac plexus. Operative time was 145' and no intra-operative adverse events were described (Fig. 4).

The postoperative course was uneventful: flatus begun the day after surgery and oral intake was restored. The patient was discharged on the 5th postoperative day. Six months later, the patient was totally free from any symptoms and a follow-up CECT shows the decompression of the celiac trunk (Fig. 5).

### 3. Discussion

The median arcuate ligament is a fibrous arch located anterior to the aorta and formed by a connection of the diaphragmatic crura. The celiac plexus is located between the arcuate ligament and the celiac trunk in up to 25% of normal individuals.

Dunbar syndrome is a rare vascular compression syndrome, characterized by postprandial intestinal angina caused by insufficient blood supply from the celiac artery to the gastrointestinal tract. Among other causes, the compression of the celiac trunk is due to a *descensus* of the diaphragm usually due to an accelerated growth in adolescents. The incidence of this disease is not known. The female to male ratio is 3:1 and the classic patient suffering from MALS is a female aged between 18 and 30 years [3–5].

Typical symptoms are chronic or recurring epigastric pain (sometimes radiating to the flanks or back), associated with vegetative symptoms including nausea, vomiting, dizziness, tachycardia, diarrhoea, weight loss and sweating; others clinical symptoms are bloating

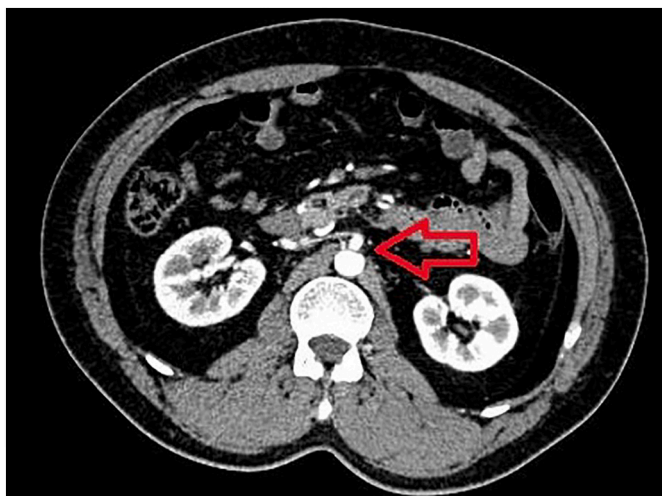


Fig. 1. Radiological features of MALS at diagnosis.

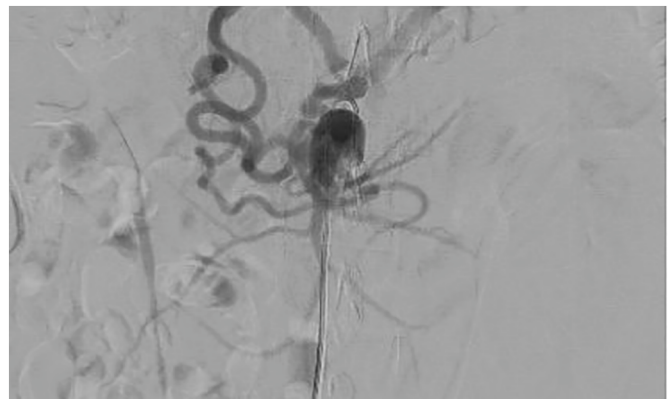


Fig. 2. Angiography showing a decreased flow from celiac trunk.

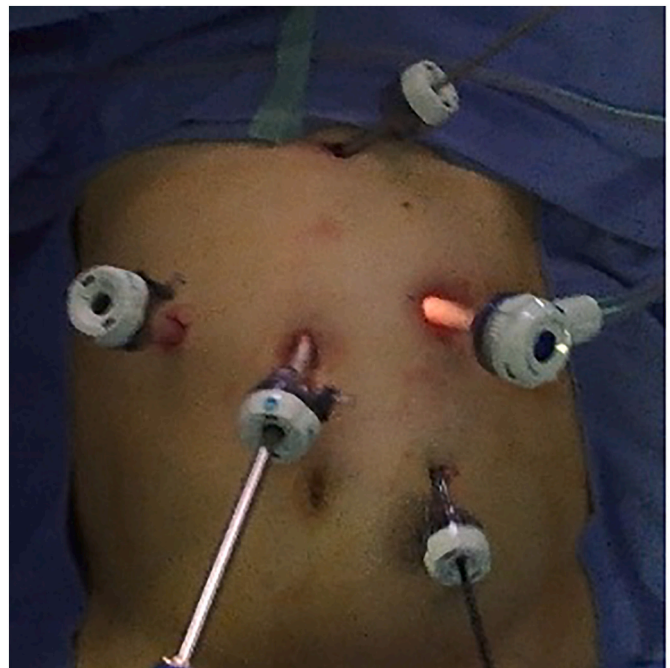


Fig. 3. Trocars' position in our laparoscopic approach.

and reduced appetite. In some cases, an epigastric bruit is detected on clinical examination [5,6]. The classical manifestation of abdominal angina is seen in about 40% of patients.

The pain seems to be related to mechanical irritation of the celiac plexus nerve fibers. Two theories have been suggested to elucidate the symptoms: a compression of the mesenteric artery producing mesenteric ischemia as well as splanchnic vasoconstriction due to stimulation of the celiac ganglion and celiac plexus [4,5].

In the present study the patient was a 40 year-old male and abdominal pain was the main symptom, related to oral intake but without weight loss.

The great challenge in Dunbar syndrome is to make a differential diagnosis: esophagitis, pancreatitis, cholelithiasis, food intolerance, mesenteric ischaemia and many other intestinal diseases may cause similar symptoms. Patients usually experiment a long-lasting history of abdominal pain and several useless medical consultations before reaching a correct diagnosis, which should be made by selective angiography, magnetic resonance angiography, spiral computed tomographic angiography and Doppler ultrasound. The combination of color duplex sonography and gastric exercise tonometry has been reported as having excellent accuracy for the diagnosis. Respiratory maneuvers are

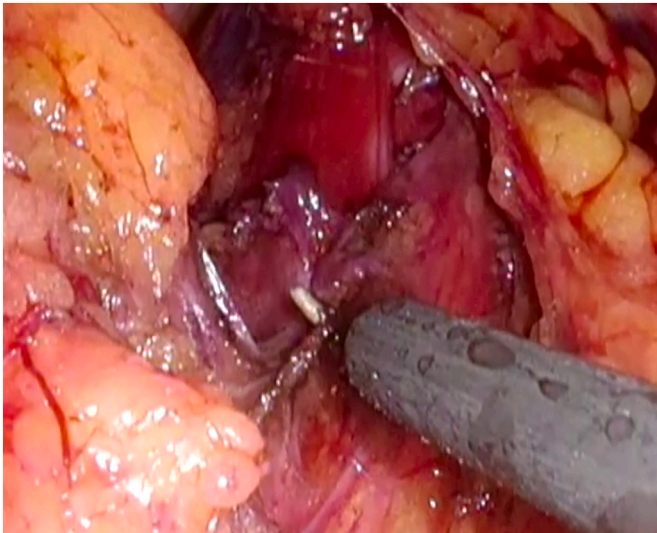


Fig. 4. Section of median arcuate ligament.



Fig. 5. CT-scan 6 months after resolutive surgery.

necessary for complete diagnosis: the demonstration of the stenosis followed by dilatation of the celiac trunk during expiration confirms the diagnosis. The typical hook-like downward displacement followed by a dilatation of the celiac artery is a typical finding [4–6]. In the present study, the diagnosis was performed by selective arteriography of the celiac trunk followed by angiotomography.

Treatment modalities in symptomatic patients include endovascular and surgical procedures (laparotomic or minimally invasive). Endovascular procedures include percutaneous transluminal angioplasty and stenting; in case of extrinsic compression release failure, surgical intervention is required [4–6]. Symptomatic patients with celiac artery compression confirmed by angiography will benefit more from surgical treatment. The operative procedure is based on the visualization and division of the arcuate ligament with decompression of the celiac artery. In selected cases a reconstruction of the artery or graft interposition is necessary. As demonstrated in literature, the minimally invasive surgery should be the treatment of choice for this condition [6–9]. The present study confirms the feasibility and safety of the laparoscopic approach for the treatment of MALS.

#### 4. Conclusion

Laparoscopic division of the arcuate ligament in patients with Dunbar syndrome is feasible and safe. In the present study, the procedure

was uneventful and restored patient's quality of life. Clear comprehension of the celiac trunk anatomy is fundamental for surgical management.

#### Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

#### Provenance and peer review

Not commissioned, externally peer-reviewed.

#### Ethical approval

In our institute, the approval of the ethics committee for the retrospective analysis of a clinical case report is not required.

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#### Guarantor

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#### Research registration number

The submitted case report is not a research study.

#### CRediT authorship contribution statement

William Sergi: design of work and manuscript writing

Norma Depalma: co-author

Stefano D'Ugo: review of surgical technique literature and authors of introduction

Ivan Botrugno, Farshad Manoochehri: data collection

Marcello Spampinato: supervisor.

#### Declaration of competing interest

The authors have nothing to disclose.

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