

# A diagnostic workup and laparoscopic approach for median arcuate ligament syndrome

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## Key words

coeliac artery, coeliac axis compression syndrome, endovascular procedures, laparoscopic surgery, median arcuate ligament syndrome.

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## Introduction

First described anatomically by Lipshutz in 1917,<sup>1</sup> median arcuate ligament syndrome (MALS) is a constellation of nonspecific symptoms attributed by a combination of intermittent ischaemic and neuropathic events. It is due to compression of the coeliac artery and entrapment of the surrounding coeliac plexus and ganglionic peri-aortic tissue. Also known as coeliac artery compression or Dunbar syndrome, symptoms are typically recurrent with nonspecific post-prandial upper abdominal pain, nausea and significant weight loss. At the level of the L1 vertebrae, the median arcuate ligament joins the diaphragmatic crura traversing superior to the coeliac axis. In 10–24% of the population, the fibrous arch of the median arcuate ligament crosses the aorta at a lower level and subsequently compresses the coeliac artery.<sup>2</sup> This may result in intermittent foregut

## Abstract

**Background:** Median arcuate ligament syndrome (MALS) is a rare disorder characterized by the compression of the coeliac trunk and plexus by fibrous arches of the median ligament. It commonly occurs in young women with postprandial epigastric pain, weight loss and nausea. We present a single surgeon experience on the diagnostic approach and management of MALS with a focus on laparoscopic surgery.

**Methods:** A retrospective review of adult patients diagnosed with MALS during a 10-year period (2011–2021) was conducted at Bankstown-Lidcombe hospital in New South Wales, Australia.

**Results:** MALS was diagnosed in six patients (mean 46 years, range: 27–74 years old), all confirmed on mesenteric duplex ultrasound and computed tomography angiography. The most common presentations were women with post-prandial pain, exercise induced pain and an average weight loss of 14.5 kg. The median interval from onset of symptoms to surgical referral was 10.5 months. The average BMI was 24.1 kg/m<sup>2</sup> and most had a grade III American Society of Anaesthesiologist physical status. All patients underwent laparoscopic release of median arcuate ligament with one patient requiring endovascular stenting. The mean operative time was 119 minutes with two minor post-operative complications, but no mortalities. The median hospital length of stay was 3.5 days with a median follow up of 3.5 years.

**Conclusion:** Laparoscopic median arcuate ligament release with endovascular support for selected cases provides sound clinical resolution of symptoms and long-term results.

ischaemia and epigastric pain. The incidence of MALS is not truly known with reports at 2 per 100,000.<sup>3</sup> It generally tends to affect younger women of low body mass.<sup>4</sup> We present a single surgeon experience for the laparoscopic management of MALS outlining a diagnostic algorithm and review of the literature.

## Methods

A retrospective review was conducted from a single surgeon's experience at Bankstown-Lidcombe hospital in New South Wales, Australia between January 2011 and July 2021. The inclusion criteria were any adult patients above the age of 18 years with chronic abdominal pain and computed tomography angiography (CTA) confirming coeliac artery compression ( $\geq 50\%$  diameter reduction of the proximal coeliac artery) and duplex mesenteric

ultrasound (US) with increased peak systolic velocities above 200 cm/s on expiration. All patients underwent thorough initial diagnostic workup to exclude other abdominal pathology including blood tests, abdominal ultrasound (US), hepatobiliary iminodiacetic acid (HIDA) scan, endoscopies (gastroscopy and colonoscopy) and cardiac review.

The data collected included:

- **Demographic information:** age, sex, body mass index<sup>5</sup> (BMI), ethnicity, smoker, comorbidities, cardiovascular disease and previous abdominal surgery.
- **Clinical presentation and symptomology:** duration of symptoms, post-prandial pain, nausea, vomiting, non-specific abdominal pain, exercise induced abdominal pain, bloating, pain worse on expiration, weight loss (kg).
- **Imaging:** Duplex US and CTA.
- **Management:** American Society of Anaesthesiologist (ASA) physical status, laparoscopic repair and/or endovascular intervention.
- **Outcomes:** operative time (hours), length of stay (LoS), post-operative complications (surgical and medical), VISICK score\*,<sup>6</sup> Gastrointestinal Symptom Rating Scale (GSRS) score\*\*,<sup>7</sup> chronic mesenteric ischaemia (CMI) score \*\*\*,<sup>8</sup> morbidity, and in-hospital mortality.

\*VISICK, a subjective comparison of pre-treatment and post-treatment symptom severity whereby patients describe their symptomatic outcome with four options (asymptomatic, improved, unchanged, and worsened). This scoring system has been previously used to evaluate the outcomes of surgical treatment of MALS.

\*\* GSRS, a 15-item survey assessing the degree of “bother” from 15 gastrointestinal symptoms on a scale of 1 (none) to 7 (very severe).

\*\*\* CMI, the score discriminates between low and high-risk patients guiding the clinical decision-making process for MALS.

Following our diagnostic workup and confirmation (Fig. 1), our collaborative approach follows a laparoscopic release of the median arcuate ligament with a vascular surgeon on stand by for review of the coeliac artery for any potential intra-operative or post-operative vascular intervention. As per the standard approach, the gastro-hepatic ligament is opened to identify the right diaphragmatic crus and the left crus concurrently identified after lateral retraction of the stomach to the left for better exposure. The distal muscular fibres of the right crus are progressively divided and dissected off using a combination of Harmonic® Scalpel (Ethicon Inc., Somerville, NJ, USA) and hook diathermy, until a good view of the anterior wall of the abdominal aorta is obtained. Similar dissection is obtained towards the left crus with generous lateral transection and retraction of the surrounding dense fibrous tissues on both sides, including the associated nerve complexes as well as lymphatics, and tracing caudally towards the origin of the coeliac trunk. The entire fibrotic bands are completely released on either side of the coeliac trunk, that is further skeletonised (Fig. 2). Quality of the decompressed artery and resultant neurolysis is ultimately reassessed conjointly with the vascular surgeon. If available, intra-operative duplex US to determine vessel patency would be ideal. A clear liquid diet is initiated on the same day with regular post-operative anti-emetics and progressed to a light diet the following day, with close monitoring

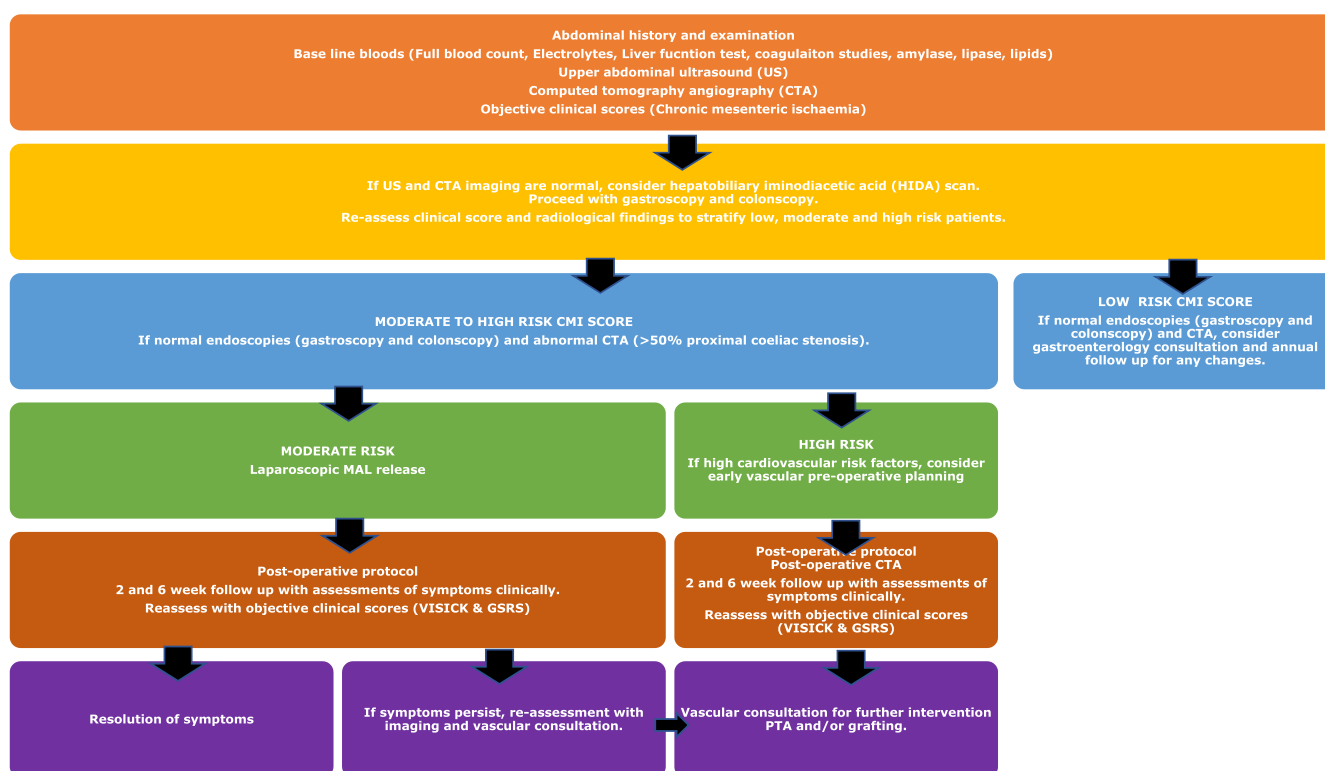
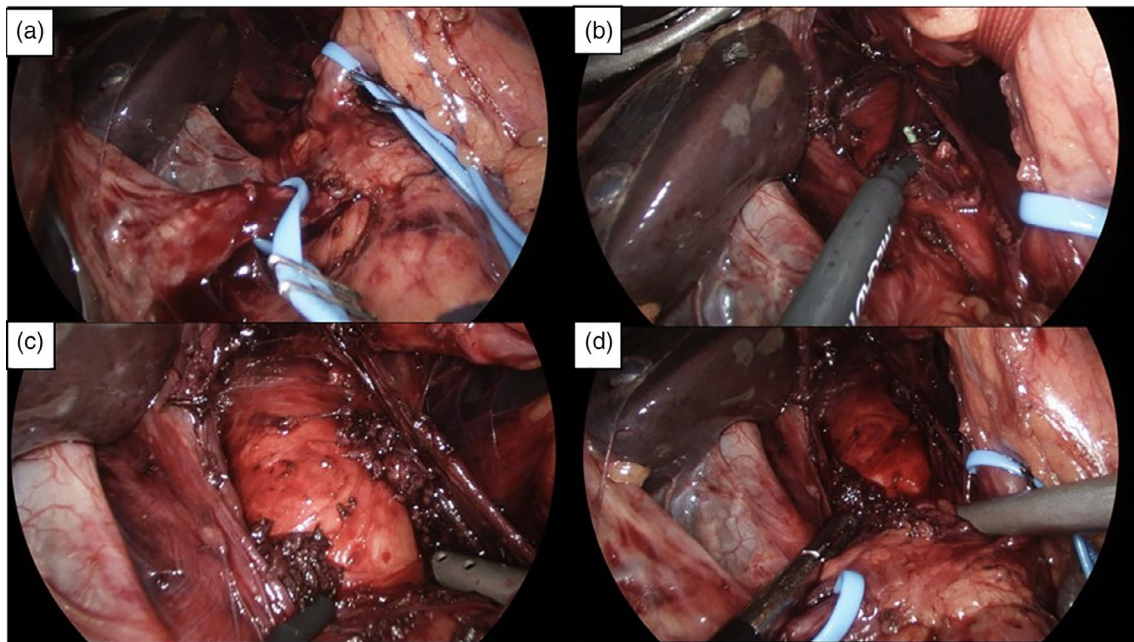


Fig. 1. Diagnostic algorithm and approach to Median Arcuate Ligament Syndrome (MALS).



**Fig. 2.** Intra-operative clinical photo for laparoscopic release of the median arcuate ligament. (a) Laparoscopic view of the inferior vena cava (IVC), caudate lobe (CL), common hepatic artery (CHA) and left gastric artery (LGA). (b and c) Progressive hook diathermy release of median arcuate ligament (CT: coeliac trunk). (d) Complete release of median arcuate ligament.

of any residual post-prandial symptoms. Routine follow-up is scheduled in clinic at 2- and 6- weeks post-operatively. Repeat CTA is recommended in moderate to high-risk cases or persistent symptoms. If residual stenosis is identified, endovascular intervention may follow. If symptoms persist after percutaneous transluminal angioplasty (PTA) and stenting, mesenteric bypass can be performed at the discretion of the vascular surgeon.

Data collected from medical records and were collated and analysed using IBM SPSS Statistics V26.0 (IBM Corp, Armonk, NY). Continuous variables were expressed as mean, median, range, and standard deviation (SD).

## Results

There were six patients (five males) with MALS with an average of 46 years old (SD = 18, range: 27–74). The average BMI was 24.1 kg/m<sup>2</sup> (range: 14.8–41.6) with two underweight and two obese. Five patients were of Caucasian ethnicity and one patient of Indian ethnicity. Half the cohort had associated cardiovascular disease and four patients had previous abdominal surgery. Two patients suffered mental health conditions including bipolar disorder and major depression. Five of six patients were smokers. The median duration of symptoms was 10.5 months (range: 3 months to 7 years, Table 1). The degree of occlusion confirmed by CTA was <50% in one case, 70% in one case, and the remaining four were ≥90%. The clinical symptoms included post-prandial pain (6/6), non-specific abdominal pain (6/6), pain worse on expiration (4/6), nausea (4/6), vomiting (3/6), bloating (3/6) and exercise induced pain (1/6). Four patients reported significant weight loss with an average of 14.5 kg (range: 5–20 kg). Most patients (4/6) had a moderate CMI score on CTA.

Five patients had a grade III ASA and one grade IV. The mean operative time was 119 min (SD±15). All patients had successful laparoscopic release of MAL and only one patient underwent further endovascular coeliac stenting 1 week post-operatively (case #6, Table 1). There were two minor complications related to one superficial wound infection treated with oral antibiotics and another of refeeding delay in a patient suffering from multiple sclerosis. Four patients reported a VISICK scores of one (asymptomatic) and two patients reported a VISICK score of two (improved) ranging from 18 months to 7 years. All patients scored a GRSR score of five. There were no reported in hospital mortalities. The median LoS was 3.5 days (range: 1–14). With a median follow up of 3.5 years (range: 7–84 months), all patients had an excellent outcome with full resolution of symptoms and no clinical recurrence.

## Discussion

MALS is a rare condition that should be considered a diagnosis of exclusion as its non-specific profile may mimic other upper gastrointestinal and abdominal pathologies. The amalgamation of both vascular and neurogenic components is more likely the origin of MALS where compression of the coeliac trunk is relieved during inspiration as the artery moves caudally, whereas compression during expiration constricts the coeliac axis in the cephalad direction.<sup>9</sup> MALS is most prevalent in young, healthy women aged between 30 and 50 years with a thin body habitus.<sup>10</sup> Consistent with the global literature, all but one of our cases was female and had an average BMI of 24.1 kg/m<sup>2</sup>. However, our experience contrasted Coehlo's findings of six patients where four had a grade I ASA score and two grade ASA II scores, respectively.<sup>11</sup> In comparison, most of our patients were overweight with grade III ASA scores;

**Table 1** Summary of clinical variables for median arcuate ligament syndrome

Patient number	Sex	Age	Cardiovascular disease	Duration of symptoms	Coeliac stenosis	SMA stenosis	ASA score	OR time (mins)	PTA	LoS	Morbidity	VISICK score	GSR score	CMI score
1	F	35	Multiple sclerosis (MS), Bipolar	3 months	50% (low)	N	3	107	N	9	MS related feeding problems	2	5	Low
2	F	54	COPD, CCF	7 years	>95% (high)	N	3	113	N	4	Superficial wound infection	1	5	Mod
3	F	52	Diabetes, Hashimoto	9 months	70% (mod)	N	3	104	N	3	-	1	5	Mod
4	F	33	CCK, HTN, dyslipidaemia	1 year	90% (high)	N	3	114	N	1	-	1	5	Mod
5	F	27	Endometriosis, HTN, dyslipidaemia	1 year	90% (high)	N	3	136	N	1	-	1	5	Mod
6	M	74	NSTEMI, dyslipidaemia, HTN, PPMI, AF	6 months	90% (high)	Y	4	140	Y	14	-	2	5	High

they also had longer median time to presentation and were marginally older (46.0 versus 43.3 years).

The most common clinical findings included post prandial pain and persistent non-specific abdominal pain, exacerbated on expiration and weight loss. Our findings support a Mayo Clinic study of 36 patients, reporting 94% with nonspecific abdominal pain, 80% postprandial abdominal pain, 50% weight loss, bloating (39%), nausea and vomiting (55.6%) and abdominal pain triggered by exercise (8%).<sup>12</sup> We reported a greater mean weight loss of 5.2 kg (14.5 versus 9.3) and proportion of obese patients compared to Huynh's experience.<sup>13</sup> These clinical presentations of rapid weight loss, pain and vomiting have been similarly reported with superior mesenteric artery syndrome (SAMS).<sup>14</sup> Although a different entity we did not identify any mixed cases of MALS and SAMS. The clinical examination is often unremarkable and non-specific with vague abdominal tenderness. Julius' findings showed that epigastric bruits were present in 16% of asymptomatic individuals and 30% of younger patients with MALS, however, we did not identify these features in our experience.<sup>15</sup>

A comprehensive workup and understanding of risk factors are central to patient selection and sound outcomes. Reilly *et al.* reported a better prognosis for patients aged between 40 and 60 years with postprandial abdominal pain, weight loss greater than 9.1 kg (20 pounds), and no history of mental illness or alcohol abuse. Similarly, Cienfuegos' study identified that patients with severe stenosis (>70%) experienced full resolution of symptoms in 71% of cases.<sup>16</sup> Brady's model further established that age and increased pre-operative expiratory coeliac artery velocity were significant predictors of clinical improvement.<sup>17</sup> In our cohort, we reported a favourable post-operative outcome for all patients with the same risk factors in conjunction with a low GSR score and VISICK score resulting in an early return to normal physical activities. Likewise, Ho reported that post-exertional pain and low VISICK scores were significantly associated with a high successful curative rate, whereas, abdominal pain at rest and vomiting along with higher VISICK scores were significantly associated with poorer outcomes.<sup>18</sup> Other risk stratification scores like CMI based on clinical and radiological features have also shown favourable outcomes to guide early clinical decision making for low, moderate, and high-risk patients (Fig. 1).<sup>19,20</sup>

As a first line of investigations, we recommend upper abdominal US along with colonoscopy and gastroscopy to exclude more common pathologies such as cholelithiasis, gastritis or pancreatitis (Fig. 1). If all are negative, we advocate duplex mesenteric US on inspiration and deep expiration with the understanding that the coeliac axis tracks cephalad during expiration, leading to external compression and elevated velocities with post-stenotic dilatation.<sup>21</sup> Findings corroborated by CTA may also demonstrate a classic hook like deformity, or post-stenotic dilation, provide CMI scoring, and concomitantly assess vasculature and other abnormalities. Magnetic resonance angiography (MRA) may also be considered as an alternative in younger patients, but due to cost and access may further delay diagnosis. Another adjunct modality may include gastric exercise tonometry to identify gastric ischaemia demonstrated by decreased gastric mucosal pH after 10-min cycling exercises.<sup>22</sup> When using digital subtraction angiography and gastric tonometry

together, Mensink reported an 86% accuracy of actual gastrointestinal ischaemia thereby optimizing surgical patient selection and benefit.

The treatment of MALS is aimed at addressing the vascular and neurogenic components of the pathology, that is, to relieve the compression of the coeliac artery restoring adequate blood flow and neurolysis. The first reports of open removal of the coeliac plexus and median arcuate ligament release were by Harjola<sup>23</sup> and Dunbar,<sup>24</sup> respectively. In 2000, Roayaie<sup>25</sup> first described laparoscopic decompression of the coeliac artery and stands as the accepted standard of surgical management with smaller incisions, decreased postoperative morbidity and improved view of the surgical field.<sup>26</sup> There is no consensus on the use of intraoperative ultrasonography with surgeons advocating for direct assessment of flow rate and those with visual inspection of median arcuate ligament release.<sup>27,28</sup> This said, intraoperative US will objectively demonstrate a return to normal peak systolic velocities.

A case series of open coeliac artery decompression reported 23 patients (50%) remained asymptomatic at follow-up, and 39 (82%) had partial or complete relief of symptoms.<sup>29</sup> We should therefore be mindful of the potential risk of recurrence after surgical release alone (24–47%).<sup>30</sup> We recommend post-operative CTA for moderate to high risk CMI patients and those with incomplete symptom relief post MAL release. Endovascular options should be discussed early with vascular surgeons to prioritize and plan for vascular reconstruction such as PTA of the coeliac artery. In more advanced cases, reimplantation of the coeliac artery on the aorta (with or without interposition grafting), and aortoceliac bypass of the stenosed segment with autologous saphenous vein or polyester graft may also be considered. For instance, our oldest patient (case #6, Table 1) had a high-grade coeliac stenosis and high risk CMI score with multiple co-morbidities and previous stents. Post-operative CTA confirmed residual stenosis of the coeliac trunk that required PTA and stenting that lead to complete resolution of residual symptoms.

Our operative time was 119 minutes, albeit slightly longer by 26 min compared to Coelho's cohort who were thinner and without previous upper abdominal operations.<sup>13</sup> Consistent with meta-analysis findings,<sup>31</sup> our protocol includes regular antiemetics, proton pump inhibitors, early mobilization and rapid progression to a light diet if tolerated. We recommend this as it reduces the possibility of post-operative nausea and vomiting seen in two of five patients in a recent series without medications.<sup>32</sup> Laparoscopic surgery for MALS has been considered as a day only procedure by some, however, our cohort had many comorbidities and travelled from regional towns resulting in a median LoS of 3.5 days seen similarly in other reports.<sup>33</sup> There were no reported mortalities, but two cases of minor complications for superficial wound infection and post-operative refeeding issues in a patient with multiple sclerosis and bipolar disorder.

In a series of 400 patients, Jimenez *et al.* reported 85% experienced immediate post-operative relief of symptoms with a rate of symptom recurrence of 6.8% in those who underwent open repair compared to 5.7% in laparoscopic release.<sup>34</sup> Roseborough and Baccari have previously demonstrated the value of laparoscopic surgery with 88% and 94% resolution over an average follow up of 28.3 and 44.2 months, respectively.<sup>35,36</sup> Both studies reported a conversion rate to open for

intraoperative bleeding in 26.7% (4/15)<sup>35</sup> and 12.5% (2/16),<sup>36</sup> respectively. Further vascular intervention was also necessary in eight cases (6/15 + 2/16) for symptom relief. With a median follow up of 3.5 years (range: 7–84 months) all patients experience excellent outcomes with no clinical recurrence or symptoms.

This study highlights the importance of the use of objective radiological and clinical measurements as adjuncts to predict successful patient outcomes, but also to anticipate those who may require vascular intervention. For trainees developing laparoscopic skills, two- and three-dimensional depth can be challenging during dissection as potential for haemorrhage or risk to the abdominal aorta from the dissection can be serious. Our experience did not report any of these complications, however, other case series have reported surgical gastrointestinal bleeding, biliary strictures, and aneurysms.<sup>37</sup> There are inherent limitations with such small case numbers, but the diagnostic approach is central to the understanding of this rare disease. Collaborative research across multiple centres would be critical in further establishing indicators for surgery and better defining clinical outcome predictors.

## Conclusion

Pre-operative workup and multidisciplinary engagement with vascular surgeons should be performed early to determine the appropriate steps of intervention on an individual basis. Laparoscopic approach combined with endovascular interventions in selected cases offers promising results with sound long-term outcomes.

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## Conflict of interest

None declared.

## Ethics statement

This project has been approved by the Human Research Ethics Committee with no restrictions. There is no source of financial or other support and no financial or professional relationships which may pose a competing interest. The data is deemed confidential and under ethics cannot be disseminated openly due to confidentiality and privacy.

## Author contributions

**Jason Diab:** Conceptualization; data curation; formal analysis; investigation; methodology; project administration; resources; software; supervision; validation; visualization; writing – original draft; writing – review and editing. **Vanessa Diab:** Data curation; formal analysis; investigation; methodology; resources; writing – original draft. **Christophe R Berney:** Conceptualization; project administration; supervision; validation; visualization; writing – review and editing.

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