

Laparoscopic Management of Celiac Artery Compression Syndrome (Dunbar Syndrome): Our Experience of 6 Cases

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DOI: <http://dx.doi.org/10.21276/ijcmsr.2019.4.2.8>

How to cite this article: Sunil Wankhade, Pravin Bhingare, Sanjay Dakhore. Laparoscopic management of celiac artery compression syndrome (dunbar syndrome): our experience of 6 cases. International Journal of Contemporary Medicine Surgery and Radiology. 2019;4(2):B36-B39.

A B S T R A C T

Introduction: Median arcuate ligament syndrome, also known as celiac artery compression syndrome or Dunbar syndrome is an uncommon condition caused by compression of celiac artery by median arcuate ligament. Its diagnosis is difficult and symptomatology is diverse. Many patients are misdiagnosed to be having inflammatory bowel disease, peptic ulcer disease and cholecystitis before a proper diagnosis is made. The suspicion of Dunbar syndrome should be aroused in any patient presenting with postprandial abdominal pain particularly if it's accompanied by weight loss. The diagnosis is usually by ultrasound and CT angiography. In patients in whom CT is contraindicated MRI can also be used for diagnosis. Management is usually done by division of median arcuate ligament and excision of celiac plexus.

Case Report: We report here our experience of 6 cases of Dunbar syndrome that were all treated by Laparoscopic resection of median arcuate ligament. Post Prandial Abdominal Pain was the cardinal symptom which was present in all the cases. The other common presenting complaints were vomiting and weight loss. On Duplex Ultrasound the mean systolic and mean end diastolic velocity of the patients was found to be 224.66 cm/s² and 59.5 cm/s² respectively. CT angiography showed characteristic focal narrowing in proximal celiac artery with its hooked appearance in all cases. All patients were successfully treated by minimal invasive laparoscopic resection of median arcuate ligament. During follow up all patients were symptoms free and had gained weight. There was no recurrence of symptoms in any of the cases treated by us.

Conclusion: Our case series shows that the celiac artery compression syndrome can be effectively treated by laparoscopic resection of median arcuate ligament.

Keywords: Dunbar Syndrome, Duplex Ultrasound, CT Angiography, Laparoscopic Resection.

INTRODUCTION

Median Arcuate ligament syndrome also known as Dunbar syndrome is a rare vascular disorder caused by compression of celiac artery by median arcuate ligament characterised by triad of Post-prandial abdominal pain, Gradual weight loss from poor food intake and vomiting.¹ In many cases the symptoms are vague and patients are labeled to be having inflammatory bowel disease, acid peptic disease and appendicitis before a proper diagnosis is made by appropriate imaging techniques. One of the most cardinal symptom which should arouse the suspicion of MAL syndrome is post prandial abdominal pain suggestive of bowel ischemia and any patient presenting with features such as postprandial abdominal pain, vomiting and weight loss must be investigated to confirm or rule out MAL syndrome. The etiopathogenesis of MAL syndrome is multifactorial and may consist of compressive effects on the celiac artery as well as on surrounding neurogenic structure (celiac ganglion). In many cases of MAL syndrome either celiac artery is located at a slightly higher level or median arcuate ligament is located at a lower level causing

compression of celiac artery and giving rise to bowel ischemia particularly in post-prandial period.²

The diagnosis can be confirmed on the basis of imaging modalities such as abdominal ultrasound, computed tomographic angiography and Magnetic resonance imaging. Ultrasonography may show presence of celiac artery narrowing at its origin from abdominal aorta. The finding of celiac artery narrowing can be further aided by Doppler examination which usually show raised systolic velocities. The ultrasonographic examination is quick, easy to perform, affordable and can be done bedside in admitted patients. But it is highly operator dependent and it has got a low sensitivity particularly in obese patients.³ For this reason CT angiography is a preferable modality of imaging in patients in whom it is not contraindicated (such as in pregnant patients). CT angiography may show narrowing of celiac artery which can be confirmed by 3D reconstructed images. It further may show extent of narrowing by indentation of compressing ligament. MRI usually is not required but may be done in co-operative patients in whom CT angiography may be contra-indicated due to radiation exposure. MRI may

show compression at the origin of celiac artery and may also identify presence of thick fibrous tissue compressing over celiac artery.⁴

Management of MAL syndrome depends upon severity and symptomatic patients are usually treated by surgical decompression. This can be performed either by open laparotomy or laparoscopic surgeries. Laparoscopic surgery is associated with less post-operative pain, fast recovery and reduced hospital stay but laparoscopic surgery needs expert surgeons and many times these surgeries may have to be converted to open surgeries due to bleeding from major vessel.⁵ Endovascular treatment by percutaneous transluminal angioplasty (PTA) and stenting is generally reserved for patients in whom surgery is contraindicated or are too old or frail for surgery. In patients treated by PTA anticoagulants are required which pose an additional risk of bleeding hence this is not recommended in young patients.⁶ We are here reporting 6 cases of MAL syndrome that were successfully treated by laparoscopic resection of median arcuate ligament. All of them were found to be asymptomatic over a follow up period of 1 year.

CASE REPORT

We here present a case series of 6 patients who were diagnosed to be having Median arcuate ligament (MAL) syndrome on the basis of imaging. All patients were treated

Age Group	No Of Patients	Percentage
< 20 yrs	1	16.67%
20-30	4	66.77%
>30	1	16.67%
Total	6	100%
Mean Age = 30.33 years		

Table-1: Age groups of the affected cases.

by laparoscopic resection of median arcuate ligament. Out of these 6 patients there were 2 males (66.67%) and 4 females (33.33%) with a M:F ratio of 1:0.5.

The study of the age group of the patients showed that MAL syndrome was usually seen in young patients. Most common affected age group was found to be between 20-30 years

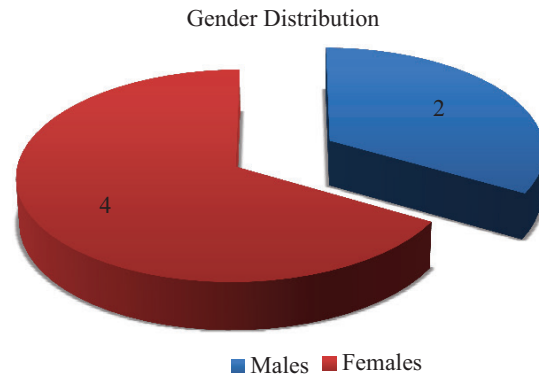


Figure-1: Gender Distribution of the studied cases.

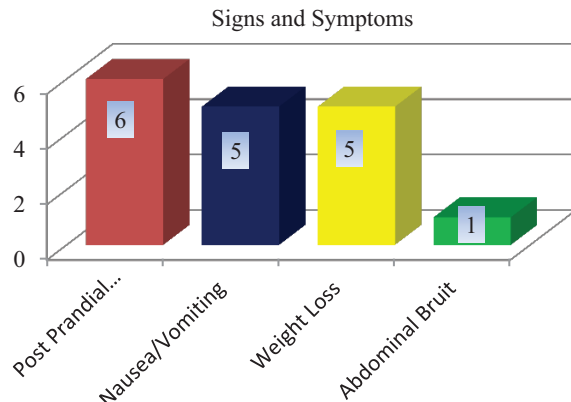


Figure-2: Signs and Symptoms in the studied cases

Sr No	Age/Sex	Symptoms	Duplex Ultra-sound (PSV) & (EDV)	CT Angiography Characteristic Focal Narrowing In Proximal Celiac Artery With Its Hooked Appearance	Intervention Minimal Invasive Laparoscopic 'MAL' Release	Follow Up (Upto 1 year)	
						Symptom Free	Wt Gain
1	30 Y/M	Post-Prandial Pain	220cm/S ² 58cm/S ²	Yes	Yes	Yes	Static
2	27 Y/F	Post-Prandial Pain Weight Loss Nausea	210cm/S ² 56cm/S ²	Yes	Yes	Yes	4 Kg
3	54 Y/F	Post-Prandial Pain Weight Loss Nausea	208cm/S ² 57cm/S ²	Yes	Yes	Yes	Static
4	30 Y/F	Post-Prandial Pain Weight Loss Nausea	210cm/S ² 58cm/S ²	Yes	Yes	Yes	2 Kg
5	22 Y/F	Post-Prandial Pain Weight Loss Nausea	240cm/S ² 60cm/S ²	Yes	Yes	Yes	2 Kg
6	19 Y /F	Post-Prandial Pain Weight Loss Nausea	260cm/S ² 68cm/S ²	Yes	Yes	Yes	4 Kg

Table-2: Demographic details, clinical features, Doppler and CT angiography features and follow up details of the studied cases.

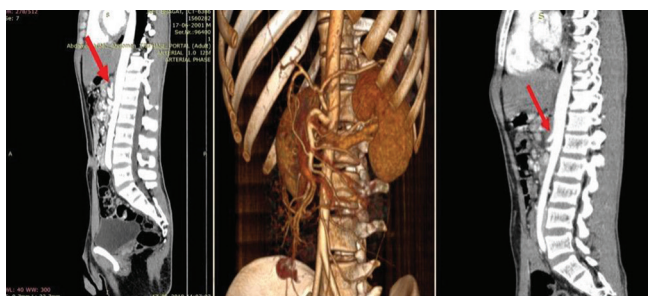


Figure-3: CT angiography sagittal view showing narrowed caliber of celiac artery along with acute angulation and indentation (Left), 3D reconstructed image showing focal narrowing, hooked appearance and post stenotic dilatation of celiac artery (Middle), Post-operative CT angiography showing normal caliber celiac artery with no evidence of indentation (Right).

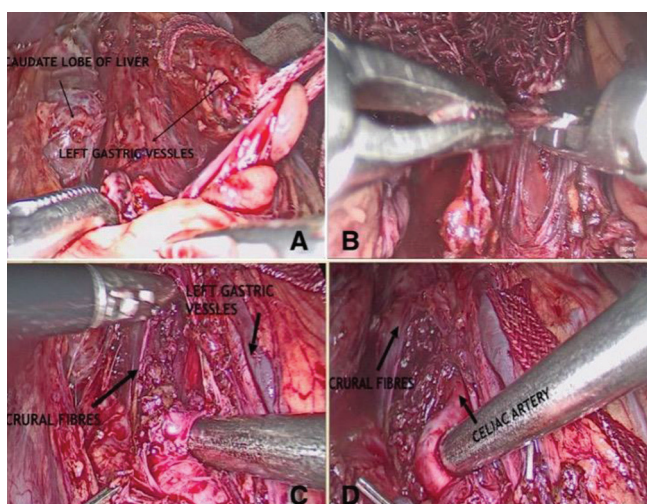


Figure-4: Technique of laparoscopic release of MAL. Left Gastric artery retracted and crural fibres are exposed (A) followed by crural dissection from caudal to cephalad (B). Intraoperative picture showing median arcuate ligament syndrome before (C) and after (D) release of median arcuate ligament.

(66.67%). The mean age of the studied cases was found to be All the patients presented with postprandial pain, weight loss, nausea and vomiting were present in 5 patients. 1 patient was found to have epigastric bruit on abdominal auscultation.

On the basis of these 3 cardinal symptoms a clinical diagnosis of bowel ischemia secondary to vascular compromise was made and imaging studies were advised. All patients underwent ultrasonography and Doppler examination. All the patients had peak systolic velocities of more than 200 cm/s^2 over the compressed segment of celiac artery. The mean systolic velocity of the patients was found to be 224.66 cm/s^2 . The mean end diastolic velocity of the studied cases was found to be 59.5 cm/s^2 .

Endoscopy was performed in all patients to assess upper gastrointestinal tract pathology. In 5 patients upper GI endoscopy was normal while 1 patient was found to be having deformed pylorus. All patients underwent CT angiography. CT angiography showed narrowing of caliber of proximal part of celiac artery. In all the cases celiac artery

caliber was found to be less than 2 mm. Mean celiac artery caliber was found to be 1.62 mm. There was acute angulation and indentation representing compression by median arcuate ligament. 3D reconstructed images clearly showed presence of focal narrowing in proximal celiac axis with distinct hooked appearance and dilatation of post stenotic part of celiac artery. Post-operative CT angiography showed resolution of indentation which was evident in preoperative CT angiography.

All patients were treated by laparoscopic release of MAL and celiac ganglion. By laparoscopic surgery MAL was cut with coagulation hook/harmonic scalpel from 10 o'clock to 2 o'clock position with nervous celiac plexus and fibrous lymphatic tissue. The end-point of surgery was complete exposure of celiac artery and freeing of celiac artery by any external stricture.

The analysis of gender, age, symptoms and imaging findings of the studied cases showed that majority of the patients were young and only 1 patient was above 50 years of age. 5 patients presented with triad of post-prandial pain, weight loss and nausea whereas 1 patient presented only with postprandial pain. The peak systolic value at celiac artery was more than 200 cm/S^2 in all the patients and CT Angiography Showed Characteristic Focal Narrowing in Proximal Celiac Artery with Its Hooked Appearance in all the cases. All patients were treated by minimally invasive laparoscopic MAL release. Median arcuate ligament was cut with coagulation hook and harmonic scalpel from 10-2 O'Clock position with nervous celiac plexus and any fibrous lymphatic tissue. Origin of celiac artery was completely exposed and freed by external stricture caused by median arcuate ligament.

During follow up period of 1 year all the patients were symptom free and there was no recurrence in any of the cases. Significant weight gain was seen in 5 out of 6 studied cases.

DISCUSSION

In 1963 Harjola described a patient who presented with postprandial abdominal pain. On Auscultation there was presence of abdominal bruit. The author reported that there was presence of thick ganglionic tissue encasing celiac artery during surgery. This thick fibrotic tissue was removed by surgery and patient was completely relieved of his symptoms. Since this first case report by Harjola many authors have reported cases of Dunbar syndrome or MAL syndrome.⁷ With advances in imaging techniques there is increased frequencies with which it is being diagnosed. Once clinically suspected the diagnosis has become pretty straight forward by CT angiography with 3D reformatting.

In our case series out of 6 cases 4 were males and 2 were females. De'Ath HD conducted a case series of 6 patients with MAL syndrome with a purpose to describe the long-term outcomes after surgical intervention.⁸ All patients with MALS who underwent laparoscopic decompression of the celiac artery were included. Surgical outcomes were recorded from a prospectively collected database. Long-term outcomes were determined by outpatient review and the Gastrointestinal Quality of Life Index (GIQLI). In this study the authors reported that out of 6 studied cases 5 were females. The median age of the studied cases was 30 years.

The female preponderance and mean age of the cases in this study was similar to the patients in our case series.

The hallmark symptoms of post-prandial abdominal pain, vomiting and weight loss are seen in majority of patients. A high index of suspicion is needed for the diagnosis and unless due consideration is given to the symptom of postprandial abdominal pain the diagnosis may be missed. An abdominal bruit is reported to be present in many patients with MAL syndrome. Sunkara T et al⁹ and Watari T et al¹⁰ and many other authors have reported presence of bruit in patients with MAL syndrome whereas bruit was absent in patients reported by Kuruvilla A et al.¹¹

The diagnosis of MAL can be suspected in young patients particularly females presenting with post-prandial abdominal pain, vomiting and weight loss. Abdominal Bruit may or may not be present. Once suspected the diagnosis can be confirmed on the basis of Imaging. Ultrasonography may show reduced caliber of celiac artery. On Doppler examination there is usually peak systolic velocity of more than 200 cm/S². In a case series of 59 cases Thomas Scholbach et al found that peak systolic velocity of more than 200 cm/s² was highly sensitive for the diagnosis of significant stenosis of celiac artery. The author reported a mean PSV of 356 cm/s² during mid-position of inspiration and expiration.¹²

CT angiography is highly sensitive and specific for the diagnosis of MAL syndrome. 3D reconstructed images not only can diagnose presence of celiac artery compression but also can show presence of collateral developed secondary to celiac artery compression. It shows characteristic hooked appearance due to indentation of celiac trunk on its superior surface. Median arcuate ligament of more than 4mm thickness on CT angiography is considered to be abnormal.¹³

The management may consist of open surgical resection, laparoscopic release and percutaneous transluminal angioplasty of the stenosed portion of celiac artery. Endovascular treatment is reserved for patients in whom surgery is contraindicated. It doesn't solve the problem and external compression and patient needs to be put on anticoagulants which poses additional risk to the patient.¹⁴ A laparoscopic surgery consists of release of compression and is associated with less post-operative pain and fast recovery.¹⁵ All patients in our case series were treated by laparoscopic approach and 5 patients remained asymptomatic during 1 year follow up period. 1 patient was lost to follow up.

CONCLUSION

A high index of suspicion is essential for proper diagnosis of patients with MAL syndrome and it should be ruled out in all the patients presenting with post-prandial abdominal pain. Laparoscopic release of median arcuate ligament is an effective treatment for symptomatic MAL syndrome. Adequate surgical release of MAL and celiac ganglion is essential for better outcome in these patients.

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Source of Support: Nil; **Conflict of Interest:** None

Submitted: 04-04-2019; **Accepted:** 26-04-2019; **Published online:** 17-05-2019