

Chapter 22

In Patients with Celiac Artery Compression Syndrome, Does Surgery Improve Quality of Life?

Grace Zee Mak

Abstract Symptomatic celiac artery compression is a controversial diagnosis that should be considered in patients with chronic abdominal pain of unknown etiology despite an extensive medical evaluation. Once suspected, patients should undergo screening mesenteric duplex. Diagnosis is confirmed with the findings of elevated celiac artery velocities which normalize with respiration followed by CT angiogram showing the typical “J-hook” conformation of the celiac artery. Patients should then undergo evaluation by a multi-disciplinary team to appropriately select patients for surgical treatment. Surgical options include release of the median arcuate ligament, with or without neurolysis of the celiac nerve plexus, and with or without concomitant revascularization procedures. Approaches can be open, laparoscopic, robotic, or retroperitoneal. Surgical treatment has an overall success rate with 70–80% patients reporting improved abdominal pain and quality of life. Post-operatively, patients can have persistent or recurrent abdominal pain and should undergo re-evaluation for possible need for revascularization procedure for stenosis of the celiac artery or celiac plexus block if the celiac artery is normalized. Additionally, some of these patients will have persistent pain consistent with a functional gastrointestinal disorder that will then require medical management.

Keywords Celiac artery compression • Median arcuate ligament syndrome • Surgical release of median arcuate ligament

G.Z. Mak, MD

Associate Professor, Department of Surgery and Pediatrics, The University of Chicago Medicine Comer Children’s Hospital, 5841 S.Maryland Avenue MC4062, A426, Chicago, IL 60637, USA

e-mail: gmak@surgery.bsd.uchicago.edu

Introduction

Celiac artery compression has been the source of much controversy since Lipshutz originally described the anatomic anomaly in 1917 followed by description of the association with digestive symptoms by Harjola (1963) and Dunbar (1965) [1, 2]. Classically, it has been described as compression at the origin by the diaphragmatic crus, or median arcuate ligament, most pronounced during expiration. This arterial compression is thought to lead to a “steal phenomenon” and foregut ischemia causing abdominal pain [1, 3, 4]. The periaortic ganglia and celiac plexus are also thought to be overstimulated leading to splanchnic vasoconstriction and ischemia further worsening the symptoms. Some postulate disruption of neuro-enteric pain pathways affecting visceral hypersensitivity mediated through the celiac ganglia [1, 3]. The classic presenting symptoms consist of post-prandial epigastric pain, nausea, and weight loss as well as the presence of an epigastric bruit increased with expiration [5]. There have also been reports of severe abdominal pain and diarrhea following exercise in well-trained athletes [3, 6, 7].

Significant controversy exists as to the true existence of this syndrome. Proponents of the syndrome attribute the symptoms to both ischemia from celiac artery compression as well as hypertrophy of the celiac nerve plexus and associated neuropathy. Histologic changes (intimal hyperplasia, elastic fiber proliferation, and disorganization of the adventitia) in the arterial wall of the celiac artery have been described in patients with celiac artery compression as well [8]. Classically, it was believed that gastrointestinal ischemia only occurred when two of the three major intestinal vessels were involved; however, many no longer support this notion and now believe that gastrointestinal ischemia is multifactorial in nature including a neurologic component [1, 4, 9]. However, opponents of the syndrome cite the incidental findings of elevated velocities and celiac artery compression with no associated symptoms as well as previous reports of inconsistent symptom improvement following surgical release [1, 4, 9].

Given this controversy, symptomatic celiac artery compression is generally considered to be a diagnosis of exclusion. Mesenteric duplex findings consist of elevated velocities of the celiac artery with normalization during deep inspiration as the ligament moves more inferiorly releasing the compression. Angiography and CT angiogram images demonstrate the classic “J-hook” conformation at the origin normalized during inspiration. With the advent of more minimally invasive, rapid, high definition, and accurate imaging modalities, this diagnosis is being made with increasing frequency [3].

Given the poorly understood pathophysiology, there has been debate regarding whether surgical treatment is indicated at all and also the exact surgery to be performed. Surgical options consist of [1] division of the ligament releasing the compression of the celiac artery at its origin [2]; with or without neurolysis of the celiac plexus, and [3] revascularization of the celiac artery either using endovascular or bypass techniques during the original procedure or at a later date. Current surgical therapy consists of open, laparoscopic, and robotic techniques. Regardless of the approach utilized, the general surgical principles are unchanged.

Many retrospective reviews have been published describing the surgical techniques and their safety, but no prospective randomized controlled studies have been performed. Thus, we can only evaluate the outcomes following surgical release of the compression and its effect on abdominal pain and overall quality of life.

Search Strategy

Literature search of English language publications was performed extending from 2006 to 2014 to identify published data on the surgical treatment of celiac artery compression in both adults and children utilizing the PICO outline shown in Table 22.1. The following databases were searched: PubMed, SUM search, and Cochrane Evidence Based Medicine.

Search words included “celiac artery compression”, “celiac artery compression syndrome”, “celiac artery compression surgery”, “celiac artery compression syndrome surgery”, “median arcuate ligament”, “median arcuate ligament syndrome”, “median arcuate ligament surgery”, and “median arcuate ligament syndrome surgery”, “Dunbar syndrome”, “Dunbar syndrome surgery”, and “celiac band compression”.

Articles not specifically addressing the surgical treatment of celiac artery compression as well as case reports of only one or two patients were excluded. There were no studies comparing operative to non-operative management in celiac artery compression, and no randomized control trials were found. Fifteen retrospective reviews and five review articles were included in the analysis. The data was classified using the GRADE system.

Results

Pre-operative Predictors of Surgical Outcome

Given the findings of asymptomatic celiac artery compression and the multitude of causes for abdominal pain, it is crucial that patients are evaluated for all possible etiologies of abdominal pain prior to being diagnosed with celiac artery compression. There has currently been very little published with specific protocols for diagnosis. Mak et al. reported the use of a specific diagnostic protocol. Complete medical evaluation should include blood work (CBC, chemistry panel, liver function tests, amylase, lipase,

Table 22.1 PICO table for surgical treatment of celiac artery compression

P (patients)	I (intervention)	C (comparator group)	O (outcomes measured)
Patients with symptomatic celiac artery compression	Surgical therapy	Patients with persistent symptoms	Relief of pain and other symptoms, Improved quality of life

erythrocyte sedimentation rate, C-reactive protein, prealbumin, thyroid function tests), upper GI, small bowel follow-through, abdominal ultrasound, upper endoscopy with biopsy, and evaluation for inflammatory bowel disease and celiac disease. Patients are then screened with mesenteric duplex. Positive findings consist of peak systolic velocities (PSV) in the celiac artery greater than 200 cm/s and an end diastolic velocity (EDV) greater than 55 cm/s. Further demonstration of a decrease in PSV with deep inspiration is suggestive of celiac artery compression. El-Hayek et al. utilized similar diagnostic criteria of PSV >200 cm/s in both inspiratory and expiratory phases [8]. Sultan et al. used these criteria as well as retrograde flow within the hepatic artery (100% predictive of severe celiac stenosis or occlusion) [1]. Patients then undergo CT angiogram to evaluate the conformation of the celiac artery in both inspiratory and expiratory phases [3].

Once the diagnosis is confirmed, it is crucial that patients are evaluated for proper patient selection for surgical intervention. Patient characteristics reported to be predictive of successful outcomes following surgery include post-prandial pain, age from 40 to 60 years, and weight loss of 10 kg or greater. Factors predictive of persistence of symptoms following surgery include atypical pain, periods of remission, age over 60 years, history of psychiatric or alcohol abuse, and weight loss of less than 10 kg [3, 6, 8, 10, 11].

Additionally, Mak et al. reported incorporating psychiatric and chronic pain service in the pre-operative and post-operative evaluations given the correlation between chronic physical pain and psychological pain. Pre-operatively, all patients are evaluated by a multi-disciplinary team consisting of general and vascular surgery, psychiatry, and pain service. This team then discusses each patient, and surgery is not considered until the patient has been unanimously cleared by the entire team [3].

Mensink described the “gastric exercise test” to detect gastrointestinal ischemia. Before, during, and after 10 min of exercise, gastric tonometry was performed measuring gastric and arterial PCO₂. Positive results were defined as gastric-arterial PCO₂ gradient >0.8 kPa after exercise, increase in gastric PCO₂ from baseline to peak exercise, and an arterial lactate level <8 mmol/L. Following surgical release of the celiac artery compression, repeat tonometry was performed at 3 and 6 months. All patients who were symptom-free post-operatively had normalized tonometry results while only 25% of patients with persistent symptoms showed normalized results. While this is not a single test that can predict success, it is an additional test to add to one’s armamentarium during the evaluation for surgical candidacy [5, 8, 12].

Surgical Technique

Published techniques for the surgical release of celiac artery compression consist of open, laparoscopic, and robotic procedures (all of which have been shown to be safe and effective). The general principles and goals of the procedures are similar – division of the median arcuate ligament including overlying lymphatics and soft tissue to release the celiac artery with or without division of the celiac nerve plexus. Some use intra-operative duplex to verify adequate release while others determine

adequate release by conformational change of the celiac artery. While there is debate regarding performance of celiac artery revascularization procedures concomitantly with the release or at a later date if symptoms recur, there is a general consensus not to perform endovascular stenting of the celiac artery pre-operatively as these stents generally fail due to external compression from the median arcuate ligament [1, 4, 5, 13]. One novel approach was described by van Petersen in which retroperitoneal endoscopic lysis of the median arcuate ligament was performed with similar safety and success rates [14].

Surgical Outcomes

The data that currently exists regarding the efficacy of surgery is quite limited with relatively short follow-up. The literature mostly consists of retrospective reviews consisting of relatively small case numbers [1–5, 7, 8, 11, 13–20]. Overall, reviews have found generally good outcomes following surgical treatment including release of the ligament (laparoscopic and open), neurolysis, and celiac artery revascularization with the majority of studies showing improved post-operative abdominal pain. Average success rate of being symptom-free following surgical intervention is reported to be 70–80% [15, 16]. Table 22.2 summarizes the findings of retrospective reviews evaluating the efficacy of surgical treatment for celiac artery compression [1–5, 7, 8, 11, 13, 14, 17–20].

One of the few larger published series by Mak et al. consists of 46 pediatric cases treated by laparoscopic release of the median arcuate ligament. The success rate was reported to be 83% with improved abdominal pain and quality of life. Post-operatively, a total of six patients required additional procedures due to persistent abdominal pain and nausea (two celiac plexus nerve blocks, two angiographies with angioplasties, one open aortoceliac bypass, and one local block at previous umbilical port incision). Of these six patients, four still reported no improvement in abdominal pain. One of the limitations of this study was the poor compliance in completing the post-operative quality of life surveys. This improved later in the study but led to poor long-term follow-up data for the initial patients [3]. The second large published series by van Petersen consisted of 46 patients who underwent retroperitoneal endoscopic release of the median arcuate ligament. They reported a success rate of 89% with 30 patients reporting no symptoms at follow-up and 11 patients reporting clear improvement of symptoms [14].

Post-operative morbidity was minimal and self-limited in the literature. Morbidity is listed in Table 22.2 [1–5, 7, 8, 11, 13, 14, 17–20]. Some patients did have self-limiting diarrhea immediately post-operatively due to the celiac sympathectomy [1].

For those patients with recurrent or persistent abdominal pain, they are re-evaluated for possible restenosis of the celiac artery either due to formation of an intravascular web or the inherent conformation of the celiac artery. These patients often require revascularization procedures either via endovascular or open approaches. Additionally, there are some patients that will have normalization of their velocities, thus indicating,

Table 22.2 Comparison of studies of operative treatment for symptomatic celiac artery compression

Study	Pts	Operative procedure performed	Post-operative results					Overall morbidity	Quality of evidence	
			Pts lost to follow-up	Complete resolution of abdominal pain	Partial resolution of abdominal pain	Overall satisfaction	Quality of life			Follow-up
El-Hayek et al. [8]	15	Laparoscopic/robotic release (2 pt underwent re-vascularization)	3	9/12 (75%)	2/12 (17%)	100%	-	15.4 months (mean)	-	Retrospective review (low)
Sultan et al. [1]	11	Open release with celiac sympathectomy (3 pt underwent re-vascularization)	0	8	1	-	-	60 months (mean)	1 (30 day) acute renal failure, chest infection	Retrospective review (low)
Cienfuegos et al. [5]	7	Laparoscopic release	-	2/3	1/3	-	-	6 months-8 years	-	Retrospective review (low)
Mak et al. [3]	46	Laparoscopic release (3 pt underwent re-vascularization)	15	31/46 (67%)	-	-	Overall improved (15/18 pt)	1.5-34.7 months	4 (fullness in chest requiring esophageal dilation in 2, pancreatitis in 1, pain at umbilical port site in 1)	Retrospective review (low)
Tulloch et al. [17]	14	Laparoscopic/open release with celiac sympathectomy (3 pt underwent re-vascularization)	0	13	-	-	-	2-65 months	1 with splenic infarction from embolization	Retrospective review (low)

Roseborough et al. [4]	15	Laparoscopic/open release with celiac sympathectomy (6 pt underwent re-vascularization)	0	9	5	–	–	–	–	–	1 with gastroparesis requiring Jtube, 1 with chronic pancreatitis	Retrospective review (low)
Kohn et al. [7]	6	Laparoscopic/open release with celiac sympathectomy	0	5 (83%)	1 (16.7%)	100%	–	–	48.6 months (mean)	0	–	Retrospective review (low)
Baccari et al. [13]	16	Laparoscopic/open release with celiac sympathectomy (2 pt underwent re-vascularization)	0	16	0	–	–	–	28.3 months (mean)	–	–	Retrospective review (low)
Berard et al. [2]	11	Laparoscopic/open release with celiac sympathectomy (1 pt underwent re-vascularization)	0	8	1	–	–	–	34 months (mean)	–	–	Retrospective review (low)
Nguyen et al. [18]	5	Laparoscopic release	0	5	0	–	–	–	1–56 months	–	–	Retrospective review (low)

(continued)

Table 22.2 (continued)

Study	Pts	Operative procedure performed	Post-operative results						Quality of evidence	
			Pts lost to follow-up	Complete resolution of abdominal pain	Partial resolution of abdominal pain	Overall satisfaction	Quality of life	Follow-up		Overall morbidity
Grottemeyer et al. [19]	18	Open release (11 pt underwent re-vascularization)	3	11 (73.33%)	-	-	-	40.68 months (mean)	Redo laparotomy in 3 pt, 2 pt with neurologic symptoms, 1 pt with pancreatitis	Retrospective review (low)
Joyce et al. [11]	6	Laparoscopic release	0	-	-	-	6	13 months (mean)	none	Retrospective review (low)
Do et al. [20]	16	Laparoscopic/robotic release with celiac sympathectomy	0	10	1	-	-	1-85 months	-	Retrospective review (low)
van Petersen et al. [14]	46	Retroperitoneal endoscopic/open release (6 pt underwent re-vascularization)	0	30 (65%)	11	-	-	2-42 months	-	Retrospective review (low)

that they have chronic functional abdominal pain. Mak et al. published a protocol for those patients with persistent symptoms. Repeat duplex ultrasound is first performed. Patients with significantly elevated velocities as well as continued respiratory variation then undergo angiography with possible angioplasty. In those patients with normalized celiac artery velocities, repeat CT angiogram is performed to evaluate for intra-abdominal pathology following surgery. If the CT is normal, patients are offered celiac plexus nerve block by anesthesia and are counseled that they may have functional abdominal pain [3]. Similar treatment algorithm was published by Duffy et al. in 2009 [21].

All the reviewed studies reported high patient satisfaction following surgical release. Though there are some patients in each study that reported no change in post-operative abdominal pain, the great majority reported at least some resolution of abdominal pain symptoms as well as overall patient satisfaction of the procedure. Even some patients who did not have complete symptomatic relief reported that they would undergo the surgery again [8].

Recommendations

Given the small case numbers and lack of randomized controlled trials, it is impossible to determine any true guidelines for diagnosis, selection of appropriate surgical candidates, or the best surgical approach. We can only develop recommendations based on the known literature.

Patients should first undergo complete medical evaluation prior to being diagnosed with celiac artery compression. Once all other diagnoses have been excluded, patients should undergo mesenteric duplex screening. If the celiac artery PSV is greater than 200 cm/s and normalizes with deep inspiration, patients should then undergo CT angiogram to evaluate the conformation of the celiac origin. Patients should then undergo evaluation by the multi-disciplinary team including general surgery, vascular surgery, psychiatry, and pain service to determine their suitability/eligibility for surgery. Patients should only undergo surgical release if they are unanimously cleared for surgery by this multi-disciplinary team. The appropriate patient selection to undergo this surgery is absolutely crucial.

Surgery should then be performed by an experienced team to ensure that the surgery is safe, adequate, and effective. The exact surgery performed can be release of the median arcuate ligament alone, combined with neurolysis, or combined with revascularization procedure. The celiac artery velocities should normalize with no respiratory variation seen on intra-operative duplex. Most patients will experience an improvement in their abdominal pain as well as overall quality of life. However, there will be a small group of patients that either do not improve or develop recurrent pain following surgery.

Initially, these patients should be re-evaluated with repeat mesenteric duplex. If the celiac velocities are elevated, they should undergo angiogram with possible angioplasty. Some patients may require multiple balloon angioplasties or even bypass reconstruction due to the formation of webs within the vessel or stenosis of

the vessel due to remodeling of the vessel from chronic compression. Those with normal celiac artery velocities should first undergo CT abdomen and pelvis to ensure the pain is not due to post-operative complications. If this is negative, they are referred to anesthesia for celiac nerve plexus blocks and may have functional GI disorder requiring medical management.

It is imperative that an algorithm be followed for not only initial evaluation of these patients but also the surgical procedure and post-operative management particularly for the later management of recurrent abdominal pain. The multi-disciplinary approach and management is extremely important throughout the entire clinical course from initial consultation to the post-operative management.

A Personal View of the Data

In patients with chronic abdominal pain of unclear etiology despite an extensive evaluation, the diagnosis of celiac artery compression syndrome should be considered. Diagnosis requires both elevated celiac artery velocities and normalization with inspiration on mesenteric duplex as well as “j-hook” conformational change seen on CT angiogram. Once diagnosed, patients should be evaluated by a multi-disciplinary team. Selecting the appropriate patients to undergo surgical treatment is absolutely crucial. Developing a plan as a multi-disciplinary team entails not only the decision to proceed with surgery but also the pre-operative preparation, immediate post-operative care, and long-term follow-up. Of critical importance is appropriately managing patient and family expectations. There should be complete candor that surgical release has been reported to be successful in 65–80% patients with improved abdominal pain and quality of life, and that there is a possibility of persistent post-operative pain.

Adequate surgical treatment requires meticulous technique to ensure complete release of the ligament so that the end result is not only a normal appearing conformation of the celiac artery from the ostia to the bifurcation but also a normalization of celiac artery velocities with no respiratory variation as well as an adequate neurectomy dividing the celiac nerve plexus. It is imperative that the surgical team has adequate general surgery and vascular expertise. The combination of minimally invasive surgical expertise as well as the vascular surgery expertise allows for a safe, effective procedure. Intra-operative duplex capabilities have allowed our group to more effectively ensure adequate lysis.

Long-term follow-up is extremely important as patients can have recurrent pain. When this occurs, it is also crucial to have a treatment algorithm including repeat mesenteric duplex, CT angiogram, angiogram with angioplasty, and bypass reconstruction. Pain that persists without increased celiac artery velocities should be treated with celiac plexus block and further treatment for functional gastro-intestinal disorders.

Randomized controlled studies of patients diagnosed with celiac artery compression comparing non-operative management to surgery or placebo surgery to surgery would be beneficial to better delineate the effectiveness of surgery; however, there are

ethical issues in the design of such trials. Additionally, our experience has been that patients seen in our clinic request surgery as they are desperate for any possible solution due to the chronic pain. Another possible study would be a randomized control trial comparing surgery alone to celiac plexus block alone to surgery with celiac plexus block. There is much opportunity to study the most effective management of these complex patients. It would also be useful to look at the patient characteristics or pre-operative evaluation that may predict success after surgical treatment as well as follow these patients for an extended period of time for long-term follow-up.

Given the small numbers of patients diagnosed with symptomatic celiac artery compression, it has been difficult to perform analysis of a large volume of patients. Thus, large multi-center studies would be necessary to perform adequate studies.

Recommendations

- In patients with celiac artery compression and chronic abdominal pain, surgeons should consider operative release of the ligament to improve pain. The success of surgical treatment for celiac artery compression is predicated on appropriate patient selection, release of the median arcuate ligament with normalization of celiac artery velocities as well as neurolysis of the celiac plexus, and appropriate post-operative follow-up with evaluation for possible need for further surgical intervention particularly in the case of recurrent abdominal pain (**evidence quality low; moderate recommendation**).
- Surgical treatment for celiac artery compression should be done only after thorough medical evaluation. The diagnosis should be confirmed by mesenteric duplex showing elevated velocities and normalization with inspiration as well as CT angiogram showing “J-hook” conformational change of the celiac artery at the origin. Additionally, there should be a multi-disciplinary pre-operative evaluation including psychiatry and pain service. There should be a frank discussion with the patient and family that this diagnosis as the cause of abdominal pain is a diagnosis of exclusion and thus the surgical treatment is a last resort. It is also imperative to manage patient expectations with the knowledge that the pain may persist following surgery (**evidence quality low; strong recommendation**).
- Patients that do not have unanimous clearance from the entire multi-disciplinary team should not undergo surgery (**evidence quality low; strong recommendation**).

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