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Resolution of Celiac Artery Compression Syndrome Following Spinal Cord Stimulation: A Case Report

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Background: Celiac artery compression syndrome is characterized by postprandial epigastric pain, weight loss, and occasional epigastric bruit, and can be difficult to treat because there are no definitive guidelines. We present a case of a 47-year-old man who had postprandial pain, weight loss, and chronic diarrhea for 5 years, which was resolved with spinal cord stimulation (SCS).

Case Report: After extensive workup, a diagnosis of celiac artery compression syndrome was confirmed. Due to previous treatment failures, repeat celiac plexus blocks becoming less efficacious, and surgical unsuitability, the patient underwent an SCS trial. They continued having pain relief, so a permanent spinal cord stimulator lead was implanted.

Conclusion: We highlight complete resolution of celiac artery compression syndrome using SCS.

Key words: Case report, celiac artery compression syndrome, celiac plexus block, median arcuate ligament syndrome, spinal cord stimulation

BACKGROUND

Celiac artery compression syndrome, also known as median arcuate ligament syndrome, is a rare disorder defined as chronic, recurrent abdominal pain related to compression of the celiac axis by the median arcuate ligament. It is clinically characterized by the triad of postprandial abdominal pain, weight loss, and abdominal bruit (1,2). The median arcuate ligament is a fibrous arch that crosses the aorta just proximal to the origin of the celiac artery and bridges the left and right crus of the diaphragm, with the celiac plexus lying within close vicinity (2). The diagnosis requires lab work and imaging to rule out more common etiologies of abdominal pain, and to confirm compression of the celiac artery by the median arcuate ligament. Caudal origin of the arcuate ligament or cranial origin of the celiac trunk can lead to compression of the celiac artery. During expiration, the diaphragm moves cranially, stretching the crura, which exacerbates celiac artery compression (2).

The diagnosis is one of exclusion with symptomatic patients presenting with a variety of nonspecific symptoms that overlap with other forms of chronic intestinal ischemia (2,3). Surgery is the only definitive treatment and options include median arcuate ligament release ± celiac ganglionectomy or surgical decompression combined with celiac artery angioplasty/stent or vascular reconstruction (4-6). We present a case in which celiac artery compression syndrome was resolved with spinal cord stimulation (SCS). This report adheres to the CARE guidelines and written, informed consent was obtained for publication.

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CASE PRESENTATION

A 47-year-old man presented with chronic, cramping abdominal pain in the mid-epigastric region over the previous 5 years. The pain was described as sharp and burning in the left-upper quadrant postprandially. Associated symptoms included chronic nausea, diarrhea, and weight loss of 30 kg over 2 years. His pain improved minimally with gabapentin, oxycodone, Lactobacillus acidophilus, probiotics, and ondansetron tablets.

Extensive testing to exclude potential etiologies, such as celiac disease, inflammatory bowel syndrome, and connective tissue, infectious, or immunologic diseases, were performed, which were either negative or fell within normal range. Initially, the patient underwent a left-upper quadrant abdominal wall rectus sheath trigger-point injection, which was ineffective.

Magnetic resonance enterorrhaphy, colonoscopy, and esophagogastroduodenoscopy results with random biopsies were negative. An ultrasound of the abdomen with color and spectral Doppler analysis were negative for ischemia. However, it did suggest significant median arcuate ligament compression on the celiac artery with elevated arterial velocities during exhalation (355 cm/s), which normalized with inspiration (158 cm/s), and patent superior mesenteric artery (SMA) and inferior mesenteric artery (IMA). Computed tomography angiogram (CTA) of the abdomen showed moderate narrowing of the celiac artery secondary to median arcuate ligament compression during expiration and was patent without poststenotic dilatation on inspiration. There was no evidence of collateral formation between celiac branches and the SMA or IMA. The vascular surgeon's impression was that although the severity of compression was atypical for celiac artery compression syndrome, he did meet other criteria with postprandial pain and significant weight loss > 9 kg. It was decided to proceed with a celiac ganglion block, and if his postprandial pain improved, he would likely benefit from median arcuate ligament release surgery.

Using the posterior transaortic approach, the first celiac plexus block was done under fluoroscopy. At his follow-up visit, the patient reported significant abdominal pain relief, both chronic and postprandial in nature, less nausea and diarrhea, and weight gain. Two successive blocks were performed and, due to the positive improvement, it was our impression that his abdominal pain represented an atypical presentation of celiac artery compression syndrome and that he would benefit from surgical intervention. However, a repeat abdominal CTA was negative for significant celiac artery compression and, while a repeat ultrasound with color and spectral Doppler did reveal an increase in the expiratory celiac artery velocity to 400 cm/s, the surgeon chose not to proceed with decompression. Over the course of a year, 5 total celiac plexus blocks were performed in identical fashion. He continued to have positive outcomes, including returning to work and improved quality of life. Unfortunately, successive blocks were proving less beneficial for the patient.

A literature review was unable to find any case reports of SCS implantation for celiac artery compression syndrome, although a case report (7) was available showing success with SCS for chronic visceral pain. Regardless, it was decided to proceed with SCS and our patient underwent placement of a temporary single SCS lead. An epidural needle was placed at T12-L1 and the lead was guided through the epidural space, so the tip of the lead was at the T4 vertebral body on fluoroscopy. Lead stimulation produced excellent coverage from the diaphragm, including both sides of the abdominal wall, and down into the legs to about the knees. At the one-week follow-up, the patient continued to have abdominal pain relief, weight gain, and was able to continue work. The patient was implanted with a permanent SCS lead within one month and continues to remain pain free. It should be noted that SCS implantation is not approved by the US Food and Drug Administration as a treatment modality for celiac artery compression syndrome.

DISCUSSION

Diagnosis of celiac artery compression syndrome is controversial and complicated by a lack of diagnostic or therapeutic guidelines (2,5). However, a diagnosis and management flow chart suggesting the use of duplex ultrasonography, angiography, and gastric exercise tonometry–if the patient presented with clinical symptoms, normal routine abdominal pain investigations, and ± incidental findings of celiac artery stenosis–can be used when patients present with incidental findings of celiac artery stenosis (5). While surgery is the only currently definitive treatment, this innovative approach of SCS implantation may benefit those patients who may not be surgical candidates (7).

Imaging studies may help identify patients with celiac artery compression syndrome (8), and celiac plexus blocks could be used to recognize patients that would respond well to SCS. Celiac plexus blocks provide symptomatic relief of celiac artery compression syndrome

that results from inflammation and compression of the celiac plexus, which serves as a relay center for abdominal visceral afferent fibers carrying pain sensation (8). This procedure involves percutaneous injection of the celiac ganglion with anesthetic agents for short-term relief. Celiac ganglion blocks have traditionally been used for both the relief of intractable pain typically associated with inoperable malignant diseases and in benign diseases, with a 73% reduction in pain from malignant diseases compared with a 37% reduction of pain from benign abdominal diseases (9). Weber et al (10) showed that 5 of 39 patients with an atypical presentation underwent a preoperative diagnostic celiac plexus block using a local anesthetic; 84.6% of these patients described symptomatic relief after the block. Initially, our patient had great pain relief with the celiac plexus block; however, successive blocks were proving less efficacious. Celiac plexus blocks could play a role in diagnosing celiac artery compression syndrome, as well as in determining if SCS will result in successful treatment. We hypothesize that positive outcomes from celiac plexus blocks may increase the success rate of SCS treating celiac artery compression syndrome.

The progressive worsening of our patient's clinical condition led us to ponder more permanent procedures, considering he was deemed an unsuitable surgical candidate. Successful treatment with SCS depends on strict criteria, and although studies or reports using SCS for celiac artery compression syndrome are lacking, there have been reports of using SCS for abdominal visceral pain. Kapural et al (7) performed SCS in 35 patients with either visceral or mixed visceral and central pain; 30 patients (86%) reported at least 50% pain relief after the trial. Due to its ischemic or neurogenic origin, compression of the celiac ganglion can lead to overstimulation of the sympathetic nervous system, and this overstimulation may be managed and controlled by SCS. We believe that the effects of SCS on celiac artery compression syndrome could be similar to those found in the treatment of other peripheral vascular diseases, possibly by decreasing sympathetic activity.

CONCLUSIONS

We reported on a patient affected by nonoperable celiac artery compression syndrome that was successfully treated with SCS. Celiac plexus blocks may play an important role in diagnosis and possible determination of successful SCS treatment of celiac artery compression syndrome. While SCS is commonly used for the management of pain secondary to ischemic vascular diseases, this successful treatment shows a need for further research. We hypothesize that positive outcomes from celiac plexus blocks may increase the success rate of SCS in treating celiac artery compression syndrome. This case highlights a successful, alternate treatment option for celiac artery compression syndrome for those not considered optimal candidates for surgery.

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REFERENCES

1. Dunbar JD, Molnar W, Beman FF, Marable SA. Compression of the celiac trunk and abdominal angina. *Am J Roentgenol Radium Ther Nucl Med* 1965; 95:731-744.
2. Scovell S, Hamdan A. Celiac artery compression syndrome. In: Eidt JF, Mills JL, Collins KA (eds). *UpToDate* 2016. Accessed 10/06/2021. www.uptodate.com/contents/celiac-artery-compression-syndrome
3. Gloviczki P, Duncan AA. Treatment of celiac artery compression syndrome: Does it really exist? *Perspect Vasc Surg Endovasc Ther* 2007; 19:259-263.
4. Duffy AJ, Panait L, Eisenberg D, Bell RL, Roberts KE, Sumpio B. Management of median arcuate ligament syndrome: A new paradigm. *Ann Vasc Surg* 2009; 23:778-784.
5. Goodall R, Langridge B, Onida S, Ellis M, Lane T, Davies AH. Median arcuate ligament syndrome. *J Vasc Surg* 2020; 71:2170-2176.
6. Takach TJ, Livesay JJ, Reul GJ Jr, Cooley DA. Celiac compression syndrome: Tailored therapy based on intraoperative findings [published correction appears in *J Am Coll Surg* 1997; 184:439]. *J Am Coll Surg* 1996; 183:606-610.
7. Kapural L, Nagem H, Tlucek H, Sessler DI. Spinal cord stimulation for chronic visceral abdominal pain. *Pain Med* 2010; 11:347-355.
8. Kim EN, Lamb K, Relles D, Moudgill N, DiMuzio PJ, Eisenberg JA. Median arcuate ligament syndrome-review of this rare disease. *JAMA Surg* 2016; 151:471-477.
9. Lee MJ, Mueller PR, vanSonnenberg E, et al. CT-Guided celiac ganglion block with alcohol. *AJR Am J Roentgenol* 1993; 161:633-636.
10. Weber JM, Boules M, Fong K, et al. Median arcuate ligament syndrome is not a vascular disease. *Ann Vasc Surg* 2016; 30:22-27.