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CASE REPORT

Celiac plexus block as a diagnostic tool in suspected pediatric median arcuate ligament syndrome

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Abstract: We report the use of celiac plexus block (CPB) as a diagnostic aid in the evaluation of median arcuate ligament syndrome (MALS) in a pediatric patient. MALS is a rare cause of severe, debilitating, abdominal postprandial pain associated with nausea, vomiting, occasional diarrhea, unintentional weight loss, and abdominal bruit. MALS is a diagnosis of exclusion, after multiple investigations. This is the first case report where CPB was used to confirm candidacy for corrective surgery in the pediatric population and has served as the cornerstone in diagnosis of MALS.

Keywords: median arcuate ligament syndrome, celiac plexus block, pediatric, diagnostic

Introduction

Median arcuate ligament syndrome (MALS) is a rare cause of chronic postprandial abdominal pain in the pediatric population. It is a diagnosis of exclusion that is suspected only after excluding other common conditions that can present with similar symptoms. One theory is that there may be a role for celiac plexus dysfunction, which can cause abnormal splanchnic vasoconstriction leading to ischemia. We present a case of the successful use of celiac plexus block (CPB) in the diagnosis of MALS in a 17-year-old patient. To our knowledge, this is the first reported case in a pediatric patient, where CPB has served as the cornerstone in diagnosis of MALS.

Case report

A 17-year-old male presented with chronic abdominal pain of 15 months duration. The patient reported that pain was more prominent in the right lower quadrant, stabbing in nature, episodic, aggravated by food intake and not relieved with antacids and proton pump inhibitors. The pain was rated as high as 10 on the numeric rating scale, which is 0–10, 0 being no pain, 10 being worst pain. Other complaints included reflux of partially digested food after eating, nausea, and vomiting, decreased oral intake, and weight loss. The patient did not have any other comorbid illnesses. He was admitted to the hospital three times for similar complaints and underwent extensive diagnostic investigations including complete blood count, complete metabolic panel, serum amylase, serum lipase, urinalysis, stool studies, *Helicobacter pylori* antigen, ELISA for Lyme disease, *Clostridium difficile* PCR, erythrocyte sedimentation rate, c-reactive protein, antinuclear antibody, SS-a and SS-b antibody, X-ray abdomen, ultrasound abdomen/pelvis, computed tomography (CT) of abdomen and pelvis, magnetic resonance enterography, capsule

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endoscopy, gastric emptying study, and Meckel's scan. All of the tests were unrevealing. Upper gastrointersinal endoscopy and colonoscopy were normal. Biopsies from esophageal, gastric, duodenal, terminal ileum, and colon were negative for organisms such as *H. pylori*, inflammation, granulomas, or viral cytopathic changes. Magnetic resonance imaging of brain was also normal, which ruled out intracranial etiologies of nausea and vomiting.

Therapeutically, he was treated with acetaminophen, ketorolac, ibuprofen, dicyclomine, and amitriptyline with little or no symptomatic relief. As there was high suspicion of appendicitis based on clinical symptoms and examination, an appendectomy was done. He was asymptomatic for ~3 months after surgery and then he started experiencing severe abdomen pain centered mainly in the epigastrium. Abdominal vascular ultrasound and CT angiography of abdomen were done. Finding suggested impression on the celiac artery by the MAL with poststenotic dilatation. He was then referred to a surgeon who specialized in MAL release for surgical management of possible MALS. However, due to patient's worsening abdomen pain and inability to tolerate food intake, he was admitted to the hospital. He was referred to inpatient pain management for diagnostic CPB to further confirm the diagnosis of MALS.

The patient was brought to the radiology suite. He was given deep sedation under monitored anesthesia care. Retrocrural two-needle approach was used under anteroposterior and lateral fluoroscopy. Correct needle position was confirmed by omnipaque contrast spread under live fluoroscopy. Subsequently, a total of 10 mL of 0.25% bupivacaine was injected on each side. As the surgeon and the patient's family wanted the block to be truly diagnostic, no steroid or additives were included in the block. Patient reported improvement of pain within 30 minutes of procedure. Pain relief lasted for 9 hours postprocedure. That evening, the patient was able to tolerate a full dinner without postprandial pain, which he had not been able to do prior to the procedure. After 9 hours, the pain returned and quickly became intolerable. He had a prolonged hospital course requiring intravenous opioids, pediatric intensive care unit stay for the severe refractory pain, placement of nasojejunal tube for feeding, and eventual discharge on sublingual morphine as needed. Given successful diagnostic block and associated clinical improvement in immediate postblock period, he was scheduled for MALS surgery

Discussion

Celiac axis is one of the three unpaired branches of abdominal aorta. It originates at the level of upper border of L1 vertebra. Anatomical variations of celiac axis and its branches have been described in the literature.[1–5] Silveira et al measured the diameter of celiac artery in 21 adult male cadavers, of which 6 showed anatomical variations in at least one of the branches. The diameter of normal celiac artery was 0.79±0.04 cm and the diameter of anatomically variable celiac artery was 0.71±0.06.[6] Even though arterial diameter in "variable" group was smaller compared to "normal" group, it did not carry any clinical significance.

Celiac artery compression at its origin by diaphragmatic crura was observed by Lipshultz in 1917.[7] The first clinical case of MALS was reported in 1963.[8] Doppler ultrasound of abdomen is used for screening and lateral aortic angiography is considered to be the gold standard investigation, in cases where MALS is suspected.[9,10] Even though 13%–50% of healthy population exhibit radiologic evidence of celiac artery compression, only a very small percentage of patients develop symptoms consistent with MALS, contributing to the controversy of its existence and validity of the current diagnostic modality.[11] Dunbar reported a series of 15 cases, which presented with postprandial abdominal pain and weight loss, and had angiographic evidence of celiac artery stenosis. Twelve out of the 15 patients had resolution of symptoms after release of celiac artery compression by MAL sectioning.[12]

Various pathophysiological mechanisms have been suggested to explain the symptoms of MALS but none has been proven. One theory is that the symptoms of MALS are due to mesenteric ischemia caused by compression of celiac artery by MAL or by the fibrotic celiac ganglion.[13] Another theory is that pain in MALS is caused by a neurogenic dysfunction resulting either directly from compression of the splanchnic nerve plexus or indirectly from splanchnic vasoconstriction.[14] In a retrospective analysis of multidetector CT angiography imaging, statistically significant correlation was demonstrated between the severity of stenosis of celiac artery and the presence of collateral circulation. The authors of this study hypothesized that ischemic complications can occur more often than was recognized by clinical signs and symptoms.[15] Some claim that postprandial pain could be the result of blood being diverted away from midgut via these collaterals resulting in a steal phenomenon.[16,17] Compression of celiac artery by MAL can also lead to aneurysms distal to the compression. Peyrottes described four cases of pancreaticoduodenal aneurysms due to MALS, of which two were ruptured and treated by endovascular repair.[18]

Surgery is often used in the treatment of symptomatic MALS. MAL release with resection of surrounding neural and lymphatic tissue is the most common procedure performed. Other less-frequent treatment options include celiac

artery bypass surgery, transluminal dilatation and angioplasty, and stent placement in refractory cases.[19]

The celiac plexus, also known as the solar plexus, is the largest plexus of the sympathetic nervous system. It relays sensory innervation of the gastrointestinal tract from stomach to splenic flexure. There are several case reports in the literature, where pain relief after CPB has been found to be a predictor of symptomatic improvement after corrective surgery.[20,21] To our knowledge, this is the first case report where CPB was used to confirm candidacy for corrective surgery in the pediatric population.

We performed the block using retrocrural two-needle approach. This approach was chosen given the comfort level of the operator, ease of access, and availability of fluoroscopy. Additional approaches include antecrural, transaortic, anterior, and endoscopic-guided approach. The authors acknowledge that there are no studies showing the superiority of one approach over the other in the setting of MALS. But each approach has its risks and benefits. In retrocrural approach, the target is the splanchnic nerves supplying the plexus and not the plexus itself.[22,23] Antecrural approach requires large volume of injectate for a successful block as the space is larger.[23,24] Transaortic approach is associated with increased risk of retroperitoneal hemorrhage, especially in patients with hypertension or coagulopathy.[22,24] Complications of anterior approach are mostly secondary to visceral organ damage.[23,24] Endoscopic approach needs a trained gastrointestinal specialist and endoscopic equipment readily available.

Manifestations of MALS include chronic postprandial abdominal pain, nausea, vomiting, occasional diarrhea, unintentional weight loss, and abdominal bruit. In a case report by Lainez et al, postprandial pain was shown to be a predictor of favorable surgical outcome in MALS.[25,26] Faries et al further demonstrated the use of gastric tonometry to determine gastric ischemia from celiac artery compression. The authors of this case report theorize that CPB may further the confirmation of diagnosis of MALS in the adult and pediatric population.

Limitations of CPB include a need for additional procedure, sedation in pediatric patients, and bleeding. The most serious complication is lower extremity complete paralysis, which occurs in <0.15% of cases. It can result from the injury to the spinal cord or damage to the anterior spinal artery. Additional less-severe neurologic complications including numbness and dysesthesia can also occur. Additional more common side effects include diarrhea and hypotension, which were not experienced in our patient.[27]

Conclusion

We present CPB as a diagnostic tool to aid in the decision for decompressive surgery in a patient with suspected MALS. Given the invasiveness and risks of surgery, after appropriate imaging is done and MALS is suspected, the authors believe that CPB may be applied as part of a diagnostic algorithm, in pediatric as well as adult patients, in the management of MALS.

Consent for publication

Written informed consent for publication was obtained from the parents of the patient.

Disclosure

The authors report no conflicts of interest in this work.

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