



Case Report

Neurolysis with Celiac Plexus Block for the Treatment of Median Arcuate Ligament Syndrome

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Citation: Shyr BS, Shyr YM, Liu CA, Chen SC, Wang SE, et al (2023) Neurolysis with Celiac Plexus Block for the Treatment of Median Arcuate Ligament Syndrome. Ann Case Report. 8: 1429. DOI:10.29011/2574-7754.101429

Received: 27 August 2023, **Accepted:** 31 August 2023, **Published:** 04 September 2023

Abstract

Median arcuate ligament syndrome (MALS) with persistent abdominal pain has long been regarded as a surgical disease and traditionally treated by surgical incision of the Median arcuate ligament (MAL) with the primary goal to restore the blood flow of celiac trunk. However, there are some observations against the role of vascular factor in the development of MALS. In this case presentation, MALS was successfully treated by percutaneous neurolysis with celiac plexus block (CPB) alone, instead of surgical MAL incision to restore the celiac arterial flow. Thus, percutaneous neurolysis with CPB can serve as not only diagnosis but also treatment of MALS.

Keywords: Celiac Plexus Block; Median Arcuate Ligament Syndrome; Neurolysis

Introduction

Median arcuate ligament syndrome (MALS) is a rare (2/100,000) entity characterized by severe, debilitating, postprandial epigastric pain associated with a constellation of other vague symptoms including nausea, vomiting, occasional diarrhea, and abdominal bruit. and/or weight loss, and even leads to an abnormal aversion to food, “sitophobia” [1,2]. The classic triad of abdominal pain related to meals, weight loss, and abdominal bruit may not always present. Typically, the diagnosis of MALS is one of exclusion, only entertained after more common conditions have been ruled out. MALS has traditionally been thought to be attributed to compression of celiac trunk, also called celiac artery compression syndrome (CACS), resulting in intestinal angina [3,4]. Therefore, surgery is often used in the treatment of symptomatic MALS. Median arcuate ligament

(MAL) release with resection of surrounding neural and lymphatic tissue to restore celiac arterial flow is the most common procedure performed [2-4]. The other theory of MALS is neuropathic, the result of median arcuate ligament impingement on the celiac plexus instead of the artery [5]. Celiac plexus block (CPB) is a radiological intervention for the treatment of visceral pain in the upper abdominal organs. It also can be used as a diagnostic tool to determine whether flank, retroperitoneal, or upper abdominal pain is sympathetically mediated via the celiac plexus or to improve prognostic accuracy before performing celiac plexus neurolysis [6]. This report is to share our experience of a successful treatment of MALS by percutaneous neurolysis with bilateral CPB, instead of surgery for MAL release.

Case presentation

This case report was approved by the Institutional Review Board of Taipei Veterans General hospital (IRB-TPEVGH No.: 2023-08-009BC). Informed consent has been obtained from the

patient for the publication of this report. A 63-year-old female presented with a 6-month history of severe chronic epigastric dull pain, which typically worsened after meal and could be alleviated by adopting a knee-to-chest position. This bothersome symptom significantly affected her daily life activities, resulting in a weight loss of 6 kg within a span of 4 months. Computed tomography-angiogram (CTA) revealed the classic “j-hook” sign, anterior indentation/compression of celiac trunk (Figure 1a). Angiography demonstrated a 2.1 cm aneurysm originating from the dilated inferior pancreaticoduodenal artery (IPDA), with retrograde contrast medium filling of the celiac branches through the dilated IPDA (Figure 1b). To prevent rupture of the IPDA aneurysm, total obliteration of the aneurysm was achieved by utilizing Nester® and Tornado® Embolization Coils (Cook Group Incorporated, Bloomington, Indiana, USA). Subsequently, a GORE® VIABAHN® Endoprosthesis stent (W. L. Gore & Associates, Inc. Flagstaff, AZ) was introduced and deployed across the aneurysm neck to seal aneurysm neck and maintain collateral flow patency to the hepatic arteries from the IPDA (Figure 1c). Neurolysis with bilateral CPB at the level of celiac trunk was performed by introducing a 22-gauge Chiba needle to the left and right celiac ganglia, which were subsequently blocked using a mixture of 4 c.c. ethanol + 4 c.c. 2% Xylocaine + 2 c.c. contrast medium (a total of 10 c.c. mixture) respectively under CT-guidance (Figure 2a-d).

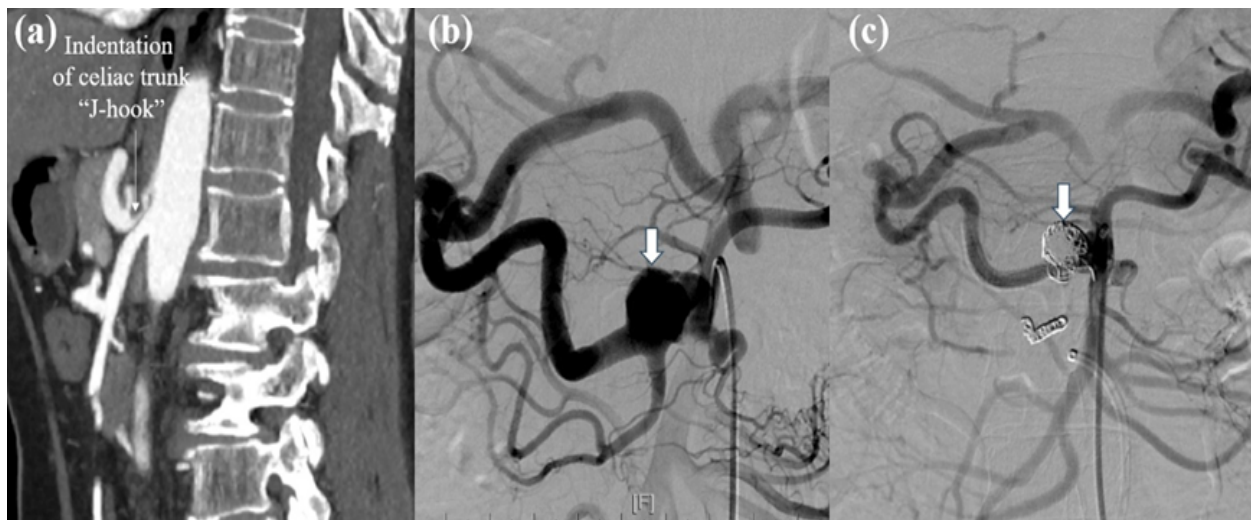


Figure 1: (a) Anterior indentation, the classic “j-hook” sign, of celiac artery on computerized tomography-angiogram (CTA), leading to the development of median arcuate ligament syndrome (MALS); (b) Presence of a 2.1 cm aneurysm (indicated by the white arrow) originating from the dilated inferior pancreaticoduodenal artery (IPDA), accompanied by retrograde contrast medium filling of the celiac branches through the dilated IPDA, resulted from the stenosis of celiac trunk; (c) Successful total obliteration (indicated by the white arrow) of the aneurysm using Nester® and Tornado® Embolization Coils, followed by a GORE® VIABAHN® Endoprosthesis stent across the aneurysm neck to seal it and maintain collateral flow patency to the hepatic arteries from the IPDA.

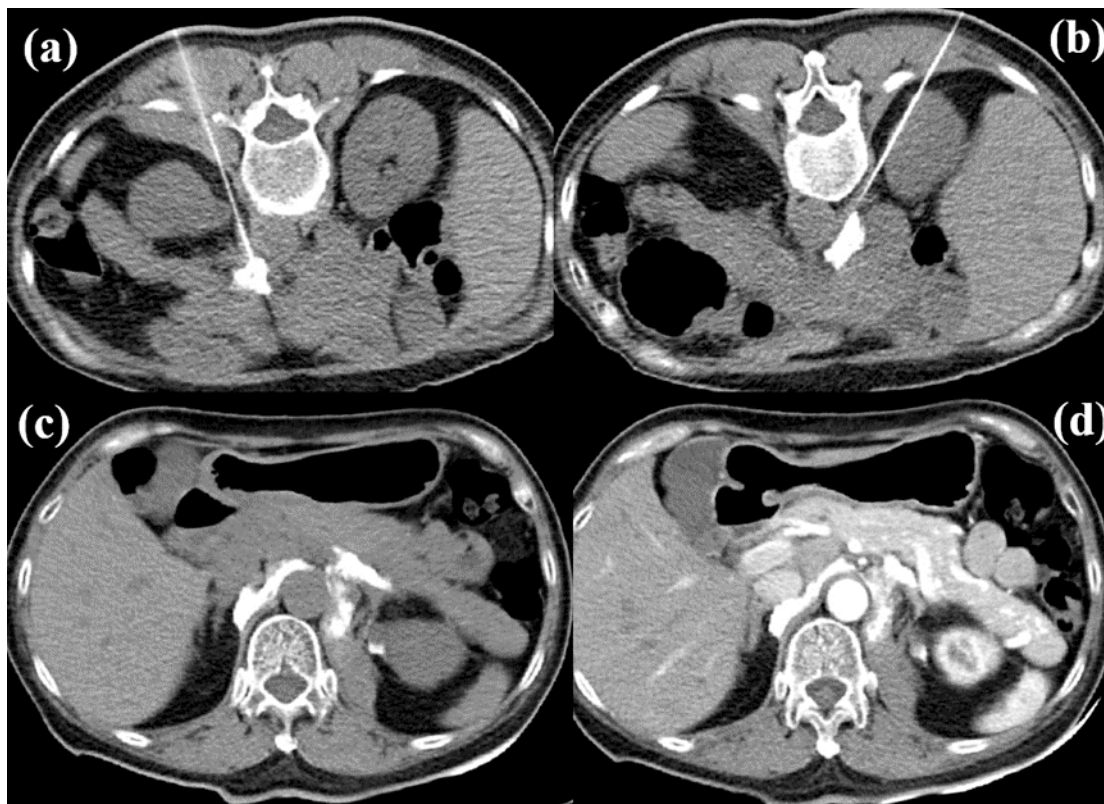


Figure 2: (a) Under computerized tomography (CT)-guidance, a 22-gauge Chiba needle was introduced to the left celiac plexus to which neurolysis was subsequently achieved by administering a mixture of 4 c.c. ethanol + 4 c.c. 2% Xylocaine + 2 c.c. contrast medium (a total of 10 c.c. mixture); (b) The same procedure was performed to block the right celiac plexus; (c) Bilateral celiac plexus block (CPB) was successfully achieved using the ethanol+xylocaine+contrast mixture, as evidenced by bilateral diffusion of the contrast on CT image; (d) The bilateral CPB at the level of celiac trunk was confirmed through a computerized tomography-angiogram (CTA) image.

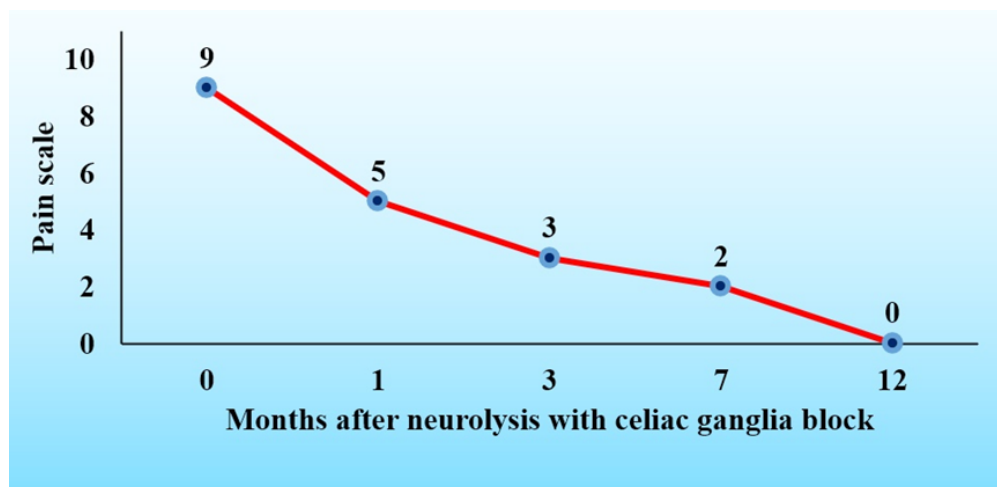


Figure 3: The Numeric Pain Rating Scale (NPRS) assessment for the patient with median arcuate ligament syndrome (MALS) following percutaneous neurolysis with bilateral celiac plexus block (CPB).

Results

Following percutaneous neurolysis with bilateral CPB, the patient experienced a remarkable improvement in the post-prandial pain related to MALS and gradually regained her body weight. One year after the procedure, she enjoyed a pain-free normal life. The Numeric Pain Rating Scale (NPRS) assessment following percutaneous neurolysis with bilateral CPB improved from 9 to 0 (Figure 3). No evidence of aneurysm recurrence was observed through imaging studies during the subsequent follow-up period.

Discussion

MALS with persistent abdominal pain has long been regarded as a surgical disease and traditionally treated with the MAL release to restore the blood flow of celiac trunk because there are no known medical therapies [7]. The arterial compression on celiac trunk in MALS is thought to lead to a “steal phenomenon” and foregut ischemia causing abdominal pain [4]. In addition, it is presumed that decreased blood flow through the celiac artery in MALS lead to development of collateral pathways in the celiac artery-superior mesenteric artery arcade and formation of aneurysms, as also seen in our case [3]. Surgical treatment has an overall success rate of 70-85% with patients reporting improved abdominal pain and quality of life [4]. Nevertheless, a study by Barbon, et al. [5] suggests that the primary etiology of MALS is neuropathic and that arterial impingement is only present in a subset of patients presenting with neuropathy. As such, the absence of celiac stenosis on imaging should not be used as an exclusion criterion for the diagnosis of MALS; conversely, the presence of celiac stenosis is not necessarily correlate with postprandial symptoms [5]. There are some observations against the role of vascular factor in the development of MALS: first, there is no report of symptoms mimicking MALS for the patients with no blood flow through celiac artery after distal pancreatectomy with “celiac axis resection”, so called DP-CAR [8]; second, even though 13% – 50% of healthy population exhibit radiologic evidence of celiac trunk compression, only a very small percentage of patients develop symptoms consistent with MALS [2,4,9]; third, only 27% of the patients with symptomatic MALS by clinical diagnosis are associated with traditional MALS anatomy with any one of the following CT findings: focal eccentric celiac artery stenosis, inferiorly displaced celiac trunk, celiac trunk hook-shape configuration, vessel collateralization or respiratory variability [5]. Given these evidences against the vascular theory in MALS and based on the fact that the celiac plexus serves as a relay center for abdominal visceral afferent fibers carrying pain sensation [6,10], it would be reasonably expected to relieve the pain by neurolysis on the celiac plexus.

Conclusion

MALS was successfully treated by percutaneous neurolysis with CPB alone, instead of surgical MAL incision to restore the

celiac arterial flow in this case presentation. Meanwhile, there was no evidence of recurrence regarding the obliterated aneurysm 1 year after transarterial embolization with Nester® and Tornado® Embolization Coils. Thus, percutaneous neurolysis with CPB can serve as not only diagnosis but also treatment of MALS.

Acknowledgements: This work was supported by grants from Taipei Veterans General Hospital (V112C-009, V112C-188, and V112B-001), the Ministry of Science and Technology (NSTC 112-2314-B-075-016), and the Ministry of Health and Welfare (MOHW111-TDU-B-221-014015).

Data availability: Data supporting this case report are available from the corresponding author on reasonable request.

Ethical Guidelines: This case report was approved by the Institutional Review Board of Taipei Veterans General hospital (IRB-TPEVGH No.: 2023-08-009BC). Informed consent has been obtained from the patient for the publication of this report.

Conflict of Interest: The authors declare that they have no conflicts of interest.

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