EISEVIER

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports



journal homepage: www.elsevier.com/locate/ijscr

Case report

Simultaneous laparoscopic treatment of median arcuate ligament syndrome and segmental adenomyomatosis of the gallbladder: A case report

Daigo Kuboki ^{a,b,*}, Ayaki Koide ^a, Satoshi Taguchi ^a, Hironobu Yamazaki ^{a,b}, Alan Kawarai Lefor ^b, Takafumi Tabuchi ^a

^a Department of Surgery, Kitaibaraki City Hospital, 1050, Sekimotoshimo, Sekinami-cho, Kitaibaraki-shi, Ibaraki, Japan

^b Department of Surgery, Division of Gastroenterological, General and Transplant Surgery, Jichi Medical University School of Medicine, 3311-1, Yakushiji, Shimotsukeshi, Tochigi, Japan

 A R T I C L E I N F O
 A B S T R A C T

 Keywords:
 Introduction: Median Arcuate Ligament Syndrome (MALS) is a condition in which the median arcuate ligament tightly compresses the celiac artery. A patient presented with MALS and segmental adenomyomatosis of the gallbladder Laparoscopic surgery Cholecystectomy

 Functional dyspepsia Case report
 Presentation of case: A 48-year-old male with adenomyomatosis of the gallbladder presented with MALS. Laparoscopic dissection of the median arcuate ligament and

artery, and the patient was diagnosed with MALS. Laparoscopic dissection of the median arcuate ligament and cholecystectomy were performed to treat both conditions. By optimizing port positions, both conditions could be treated simultaneously. The patient was discharged on postoperative day 6 and has no recurrent symptoms 20 months postoperatively.

Discussion: To the best of our knowledge, there are no previously reported cases of simultaneous laparoscopic division of the median arcuate ligament and cholecystectomy.

Conclusion: By optimizing the port positions, laparoscopic division of the median arcuate ligament and cholecystectomy were performed simultaneously, minimally invasively, safely and effectively.

1. Introduction

Median arcuate ligament syndrome (MALS) is a rare condition in which a fibrous diaphragmatic arcuate ligament causes compression of the celiac artery and/or the celiac plexus nerves. Extrinsic compression of the celiac artery can lead to abdominal pain postprandially or during expiration. However, postprandial epigastric pain is often treated as functional abdominal pain and may progress without improvement [1]. Here, we report a patient with MALS diagnosed on the basis of postprandial epigastric pain which was initially treated as functional dyspepsia but did not improve. Laparoscopic dissection of the median arcuate ligament to treat MALS and cholecystectomy to treat gallbladder adenomyomatosis were performed simultaneously. To the best of our knowledge, there are no previously reported cases of simultaneous laparoscopic division of the median arcuate ligament and cholecystectomy. This report was prepared in compliance with the SCARE criteria [2].

2. Presentation of case

A 48-year-old man was diagnosed with functional dyspepsia based on postprandial epigastric pain and was treated medically. Four months later, without improvement, the postprandial epigastric pain became more intense, and the patient presented to our facility. Height, weight, and BMI were 169.5 cm, 47.5 kg and 16.5 kg/m². Physical examination of the abdomen was soft and flat, no tenderness, no palpable masses and normal bowel sounds. Laboratory tests were within normal limit. An upper endoscopy was performed and was normal. Enhanced threedimensional computed tomography (3D-CT) scan of the abdomen showed compression of the celiac artery (Fig. 1a) and diffuse wall thickening from the body to the fundus of the gallbladder (Fig. 1b). Abdominal ultrasound imaging showed deformity of the celiac artery, and abdominal Doppler ultrasound imaging showed that the blood flow in the celiac artery varied during the respiratory cycle. In addition, abdominal ultrasound showed thickening of the gallbladder wall from the body to the fundus and Rokitansky-Aschoff sinuses were scattered in

https://doi.org/10.1016/j.ijscr.2022.107869

Received 2 October 2022; Received in revised form 29 December 2022; Accepted 29 December 2022 Available online 2 January 2023

^{*} Corresponding author at: Jichi Medical University School of Medicine, 3311-1, Yakushiji, Shimotsuke-shi, Tochigi, Japan. *E-mail address*: m05037dk@jichi.ac.jp (D. Kuboki).

^{2210-2612/© 2023} The Authors. Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

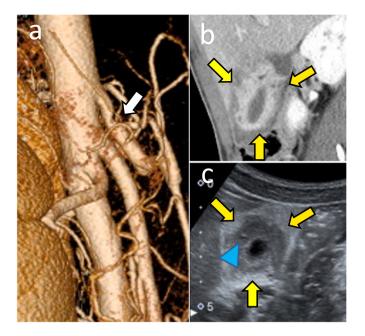


Fig. 1. a. Preoperative three-dimensional computed tomography angiography showed compression of the celiac artery (white arrow). b. Preoperative enhanced computed tomography scan showed diffuse wall thickening from the body to the fundus of the gallbladder (yellow arrows). c. Preoperative abdominal ultrasonography imaging showed thickening of the gallbladder wall from the body to the fundus (yellow arrows) and Rokitansky-Aschoff sinuses were scattered in the wall (blue arrowhead). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

the wall (Fig. 1c). The patient was diagnosed with MALS and segmental ademyomatosis of the gallbladder. The patient had previously been advised to undergo surgery for adenomyomatosis of the gallbladder but declined at that time. The patient was now diagnosed with MALS with postprandial pain and desired surgery for MALS. It was recommended that a cholecystectomy be performed simultaneously, and the patient agreed. After obtaining informed consent, laparoscopic ligament dissection and cholecystectomy were planned.

2.1. Operative procedure

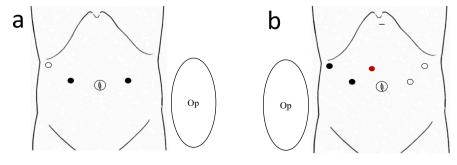
Surgery was performed under general anesthesia in the supine position. The open method was used to insert the 12-mm umbilical camera port. Following carbon dioxide insufflation, two 5-mm ports were placed in the right upper and lateral abdomen, and a 5-mm port in the left lateral abdomen. Insufflation was maintained at 10 mmHg pressure. Laparoscopy was performed with a flexible laparoscope (Olympus, Tokyo, Japan). The operator stood on the left side of the patient (Fig. 2a). First, cholecystectomy was performed in the usual manner.

The specimen was removed through the umbilical port wound using an ENDO CATCH™ II (Covidien Japan, Tokyo, Japan). Next, the operator moved to the right side of the patient to perform median arcuate ligament dissection. The patient was placed flat in a reverse Trendelenburg position. An additional 5-mm port was placed in the left upper abdomen, and a Nathanson Hook Liver Retractor™ (Yufu Itonaga Co.,Ltd., Tokyo, Japan) was inserted through the epigastric area to lift the left hepatic lobe (Fig. 2b). LigaSure[™] Maryland (Covidien Japan, Tokyo, Japan) was used to open the minor omentum and dissect between the right diaphragmatic crus and the gastric ligament. The left gastric coronary vein was clipped and divided, and the left gastric artery was retracted to the left using a 6-mm Penrose drain (Fuji Systems Corporation, Tokyo, Japan) (Fig. 3a). After the retroperitoneum was dissected, the left and right diaphragmatic crura were separated, and the celiac artery was identified (Fig. 3b). Laparoscopic ultrasonography was used to visualize and confirm the compression of the celiac artery. Because of the difficulty in dissecting the anterior surface of the celiac artery, an additional 5-mm port was inserted to the right and superior to the umbilicus. After exposing the anterior surface of the celiac artery, thick white connective tissue that ran laterally was observed, and the MAL was identified (Fig. 3c). When it was divided, the origin of the celiac artery was exposed, and the compression of the celiac artery had been released (Fig. 3d). Intraoperative Doppler ultrasonography confirmed that the celiac artery was no longer compressed, and the blood flow improved. The operating time was 229 min, with minimal blood loss. After surgery, the patient's symptoms were relieved, and decompression of the celiac artery was confirmed using 3D-CT scan (Fig. 4). The patient was discharged on postoperative day 6 and has no recurrent symptoms at 20 months postoperatively. Histopathological examination of the gallbladder showed only gallbladder adenomyomatosis, with no evidence of malignancy.

3. Discussion

The median arcuate ligament is a band of fibrous tissue that anteriorly connects the diaphragmatic crura surrounding the aortic hiatus. MALS was first described in 1963 as a syndrome attributed to compression of the celiac artery by the median arcuate ligament [3], and the first surgical series was reported by Dunbar in 1965 [4]. Although several case series were reported, the true incidence of MALS remains unknown. Compression of the celiac artery by the median arcuate ligament is present in 3.4-7.3 % of asymptomatic patients in whom imaging is performed for other indications [5]. Typical symptoms include postprandial epigastric pain, nausea or vomiting, and weight loss, but most patients with compression of the celiac artery due to the median arcuate ligament are asymptomatic. Only approximately 1 % of those with median arcuate ligament compression develop MALS [6] and few patients require treatment. Recently, MALS has attracted interest in relation to pancreatoduodenal arcade aneurysms. In MALS, the superior mesenteric artery "steals" blood flow from the common hepatic artery resulting in visceral ischemia (the "steal" phenomenon) that is aggravated post-prandially. Reversal of the direction of flow in the common

Fig. 2. a. Port sites during laparoscopic cholecystectomy. A 12-mm umbilical camera port, 5-mm ports in the right upper and lateral abdomen, and the left lateral abdomen were inserted. The operator stood on the left side of the patient. The ports used by the operator are shown in black. Op: Operator. b. Port sites during the median arcuate ligament dissection. A 5-mm port was added in the left upper abdomen, and a liver retractor was inserted at the epigastrium. The operator stood on the right side of the patient. A 5-mm port was added on the right side of the patient. A 5-mm port was added on the right side for the operator (red). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)



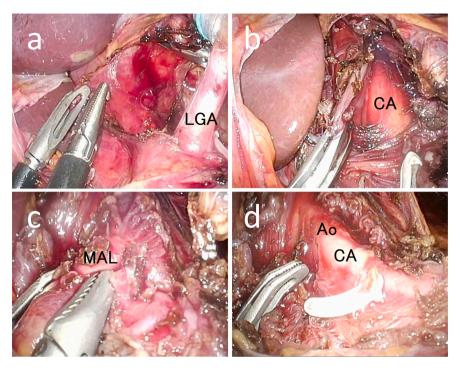


Fig. 3. a. Surgical procedure for median arcuate ligament syndrome. The left gastric coronary vein was clipped and divided, and the left gastric artery was retracted to the left. b. The left and right diaphragmatic crura were separated, and the celiac artery wall was confirmed. c. The median arcuate ligament was identified. d. After dissection of the median arcuate ligament, the origin of the celiac artery was exposed, and compression of the celiac artery, CA; celiac artery, MAL; median arcuate ligament, Ao; aorta.

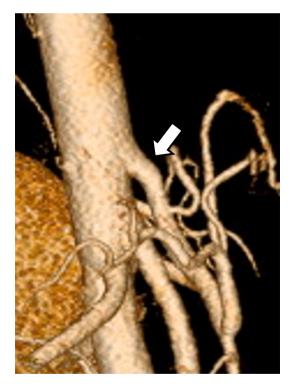


Fig. 4. Postoperative three-dimensional computed tomography angiography showed that compression of the celiac artery was released (arrow).

hepatic artery may result in a pancreatoduodenal arcade aneurysm due to elevated intravascular pressure [7]. The probability of rupture is much higher for a pancreaticoduodenal arcade aneurysm than for other aneurysms because a pancreaticoduodenal arcade aneurysm tends to rupture regardless of size. Therefore, once a pancreatoduodenal arcade aneurysm is diagnosed, it should be treated promptly as ruptured aneurysms are life-threatening [8].

In addition, one of the characteristics of MALS is that the associated

symptoms are usually nonspecific and are easily misdiagnosed as functional abdominal pain such as functional dyspepsia, peptic ulcer disease, or gastropathy [1] [9]. In the present patient, postprandial epigastric pain was initially treated as functional dyspepsia, and it took time to diagnose MALS. Noticing a compressed celiac artery on 3D-CT scan can facilitate making the correct diagnosis.

MALS is treated by decompression of the celiac artery [3] [4]. Until recently, open surgery was the therapy of choice. Recently, reports of laparoscopic treatment have become more common. Laparoscopic treatment of MALS compared with open surgery has several advantages, including less morbidity, less postoperative pain, shorter recovery period, less adhesions, less blood loss, earlier resumption of oral intake, faster return to normal activities, shorter hospital stay and better cosmetic results [5] [10] [11] [12]. Jimenez et al. showed that late recurrence rates were similar after laparoscopic versus open surgery [13]. In addition, there are also reports of robot-assisted repair of MALS [14].

To the best of our knowledge, there are no previous reports of a pathophysiologic link between MALS and adenomyomatosis of the gallbladder. It is reported that adenomyomatosis of the gallbladder also causes abdominal pain [15]. Therefore, both MALS and adenomyomatosis of the gallbladder could have been causes of pain in this patient. If surgery was performed for only one of the conditions, there was a possibility of residual symptoms, and the patient desired surgery for both conditions simultaneously. In addition, segmental adenomyomatosis of the gallbladder is a premalignant condition, and resection was deemed appropriate. Intestinal ischemia and decreased intestinal peristalsis due to compression of the celiac artery may result in the formation of gallbladder stones.

In the present patient, laparoscopic division of the median arcuate ligament and cholecystectomy were performed simultaneously. Although there are many reports of laparoscopic division of the median arcuate ligament, to the best of our knowledge, there are no reported cases in which cholecystectomy was performed simultaneously. Performing these two operations simultaneously required careful consideration of port placement. The port positions were decided based on the ports used for laparoscopic gastrectomy, because cholecystectomy is often performed during gastrectomy. Placing the right upper abdominal

port in a slightly lateral position made it easier to elevate the gallbladder. During median arcuate ligament dissection, five ports and liver retraction were used. During dissection of the connective tissue along the anterior wall of the celiac artery, the right-hand forceps was tangential which made dissection difficult. Another port was placed at a level superior to the umbilicus, which facilitated safe dissection with minimal bleeding. There are several reports describing port positioning for median arcuate ligament dissection, similar to the positioning for gastrectomy [16] [17]. However, MALS requires dissection along the anterior surface of the celiac artery, which is deeper than during a typical gastrectomy. Therefore, the pancreas may overhang and interfere with the path of the forceps (Fig. 5). In some cases, the working ports were placed cephalad from the beginning [18]. We believe it is important not to hesitate to add a port to perform the procedure safely. Tulloch et al. surveyed 14 patients with MALS and found that "Median operative times were 220 min (160-280 min) in the laparoscopic surgery group and 245.5 min (150-350 min) in the open surgery group." Laparoscopic surgery tended to have shorter operative times than open surgery [5]. Considering that a cholecystectomy was also performed, an operative time of 229 min in this case is reasonable.

In summary, MALS in a patient with gallbladder adenomyomatosis was diagnosed by 3D-CT scan and Doppler ultrasonography which was originally diagnosed as functional dyspepsia and treated without improvement. By optimizing the port positions, it was possible to perform laparoscopic release of the median arcuate ligament and cholecystectomy at the same time. Both procedures were performed safely, the postoperative wound gave an excellent cosmetic result, and the symptoms have not recurred.

4. Conclusion

Laparoscopic cholecystectomy and median arcuate ligament dissection were performed simultaneously, minimally, invasively, safely and effectively by optimizing the port positions.

CRediT authorship contribution statement

Daigo Kuboki and Ayaki Koide performed the surgery.

All authors performed acquisition of data, analysis and interpretation of data.

Daigo Kuboki, Alan Kawarai Lefor and Takafumi Tabuchi performed drafting the article and revising it critically for important intellectual content.

All authors read and approved the final manuscript.

Consent

Written informed consent was obtained from the patient for the publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

Ethic approval is exempt at our institution.

Sources of funding

N/A

Research registration

N/A

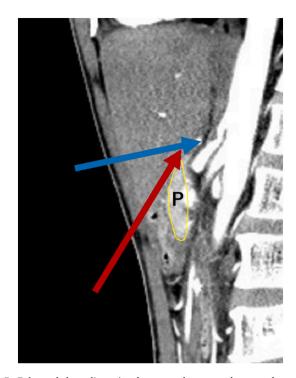


Fig. 5. Enhanced three-dimensional computed tomography scan showed the location of celiac artery and pancreas. Forceps from the caudal side was obstructed by the pancreas (red arrow).

Forceps from the temporal side of the umbilicus (additional port) reached the celiac artery directly (blue arrow). P; Pancreas. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

Guarantor

Daigo Kuboki is a guarantor of this study.

Declaration of competing interest

N/A

Acknowledgements

I would like to express my appreciation to Akira Saito for his thoughtful guidance and support in writing the manuscript and technical editing.

References

- M. Scharf, K.A. Thomas, N. Sundaram, S.J.K. Ravi, M. Aman, Median arcuate ligament syndrome masquerading as functional abdominal pain syndrome, Cureus 13 (2021), e20573.
- [2] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, A. Kerwan, The SCARE 2020 guideline: updating consensus surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230.
- [3] P.T. Harjola, A rare obstruction of the coeliac artery. Report of a CASE, Ann. Chir. Gynaecol. Fenn. 52 (1963) 547–550.
- [4] J.D. Dunbar, W. Molnar, F.F. Beman, S.A. Marable, Compression of the celiac trunk and abdominal angina, Am. J. Roentgenol. Radium Therapy, Nucl. Med. 95 (1965) 731–744.
- [5] A.W. Tulloch, J.C. Jimenez, P.F. Lawrence, E.P. Dutson, W.S. Moore, D.A. Rigberg, et al., Laparoscopic versus open celiac ganglionectomy in patients with median arcuate ligament syndrome, J. Vasc. Surg. 52 (2010) 1283–1289.
- [6] K.M. Horton, M.A. Talamini, E.K. Fishman, Median arcuate ligament syndrome: evaluation with CT angiography, Radiographics 25 (2005) 1177–1182.
- [7] K. Matsumoto, H. Shinozaki, S. Shinozaki, T. Terauchi, A.K. Lefor, N. Sata, Normalization of flow in the common hepatic artery after decompression of median arcuate ligament syndrome with diminution of a pancreatoduodenal arcade aneurysm, Case Rep. Gastroenterol. 16 (2022) 284–289.

D. Kuboki et al.

International Journal of Surgery Case Reports 103 (2023) 107869

- [8] T. Terayama, Y. Tanaka, S. Soga, H. Tsujimoto, Y. Yoshimura, Y. Sekine, et al., The benefits of extrinsic ligament release for potentially hemodynamically unstable pancreaticoduodenal arcade aneurysm with median arcuate ligament syndrome: a case report, BMC Surg. 19 (2019) 50.
- [9] J.A. Cienfuegos, F. Rotellar, V. Valentí, J. Arredondo, N. Pedano, A. Bueno, et al., The celiac axis compression syndrome (CACS): critical review in the laparoscopic era, Rev. Esp. Enferm. Dig. 102 (2010) 193–201.
- [10] J.C.U. Coelho, A.V.E. Hosni, C.M. Claus, Y.S.H. Aguilera, G.P. Abot, A.T.C. Freitas, et al., Treatment of median arcuate ligament syndrome: outcome of laparoscopic approach, Arq. Bras. Cir. Dig. 33 (2020), e1495.
- [11] E.N. Kim, K. Lamb, D. Relles, N. Moudgill, P.J. DiMuzio, J.A. Eisenberg, Median arcuate ligament syndrome-review of this rare disease, JAMA Surg. 151 (2016) 471–477.
- [12] H.D. De'Ath, S. Wong, K. Szentpali, S. Somers, T. Peck, C.H. Wakefield, The laparoscopic management of median arcuate ligament syndrome and its long-term outcomes, J. Laparoendosc. Adv. Surg. Tech. A 28 (2018) 1359–1363.

- [13] J.C. Jimenez, M. Harlander-Locke, E.P. Dutson, Open and laparoscopic treatment of median arcuate ligament syndrome, J. Vasc. Surg. 56 (2012) 869–873.
- [14] C. Fernstrum, M. Pryor, G.P. Wright, A.M. Wolf, Robotic surgery for median arcuate ligament syndrome, Jsls (2020) 24.
- [15] S. Teelucksingh, T. Welch, A. Chan, J. Diljohn, F.S. Rampersad, Gallbladder adenomyomatosis presenting with abdominal pain, Cureus 12 (2020), e10485.
- [16] Y. Fujiwara, M. Higashida, H. Kubota, Y. Watanabe, M. Ueno, M. Uraoka, et al., Laparoscopic treatment of median arcuate ligament syndrome in a 16-year-old male, Int. J. Surg. Case Rep. 52 (2018) 79–83.
- [17] J. Okadome, J. Okazaki, E. Kawakubo, R. Kyuragi, K. Homma, A. Guntani, et al., A case of median arcuate ligament syndrome managed by laparoscopic surgery and additional endovascular treatment: report of a case, J. Jpn. College Surg. 40 (2015) 683–686.
- [18] S. Gülmez, U. Aday, A.S. Senger, E. Gündeş, A new laparoscopic manoeuvre in median arcuate ligament syndrome, Balkan Med. J. 34 (2017) 590–592.