



Citation: Malerba, S., Puglisi, G.R., Panzera, P., Pasculli, A., Prete, F.P., Gurrado, A., & Testini, M. (2023). Wandering gallbladder wrapping hepatoduodenal ligament and mimicking gastrointestinal stromal tumor at imaging. Case report and literature review. *Italian Journal of Anatomy and Embryology* 127(2): 13-17. doi:10.36253/ijae-14548

Copyright: © 2023 Malerba, S., Puglisi, G.R., Panzera, P., Pasculli, A., Prete, F.P., Gurrado, A., & Testini, M. This is an open access, peer-reviewed article published by Firenze University Press (<http://www.fupress.com/ijae>) and distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited.

Data Availability Statement: All relevant data are within the paper and its Supporting Information files.

Competing Interests: The Author(s) declare(s) no conflict of interest.

Wandering gallbladder wrapping hepatoduodenal ligament and mimicking gastrointestinal stromal tumor at imaging. Case report and literature review

SILVIA MALERBA, GIULIANA RACHELE PUGLISI, PIERCARMINE PANZERA, ALESSANDRO PASCULLI, FRANCESCO PAOLO PRETE, ANGELA GURRADO, MARIO TESTINI*

Department of Precision and Regenerative Medicine and Ionian Area. University of Bari, Italy

*Corresponding author. E-mail: mario.testini@uniba.it

The first two authors contributed equally to the work

Abstract. Wandering or free-floating gallbladder is a rare anomaly of position of the gallbladder, characterized by an unusually long or absent mesentery. This condition may present clinically as an emergency case of gallbladder torsion or as part of a complex clinical picture, where patients may present symptoms that trace to gallbladder pathology, but show morphologic features that are distant from gallbladder disease. We present a case where a wandering gallbladder wrapping around the hepatoduodenal ligament adhered to the stomach mimicking features of a gastrointestinal stromal tumor at preoperative imaging.

Keywords: ectopic gallbladder, cholecystectomy, anatomical variant, position anomaly; gastrointestinal stromal tumor.

INTRODUCTION

Cholelithiasis is one of the most common surgical diseases; its estimated overall prevalence is 10–15% in the general population, with some differences across Countries, and the majority of patients are asymptomatic [1, 2].

Laparoscopic cholecystectomy (LC) is considered worldwide the gold standard for the treatment of symptomatic gallbladder lithiasis, and it is currently one of the most frequently performed procedures in general surgery [3]. However, this procedure has rare but potentially very serious complications[4].

Guidelines and technical suggestions have been published to offer indications for a safe cholecystectomy, although this body of evidence most commonly refers to the normal anatomical situation of the gallbladder with its most common variants[5]. Some anomalies of the gallbladder position may present unexpected challenges to the surgeon but also to the physician investigating symptoms that may involve gallbladder pathology.

There are different types of anomalies and anatomic variants of gallbladder: agenesis, duplication, anomalies of shape, of size, and position. For what concerns the defects of position, they are divided in wandering gallbladder, gallbladder torsion and ectopic gallbladder. An ectopic gallbladder usually can be intrahepatic, suprahepatic, retrohepatic, supradiaphragmatic, and retroperitoneal; left-sided gallbladders may occur in *situs inversus* or as an isolated finding [6]. Ectopic gallbladder can also lie in the falciform ligament, transverse mesocolon, and anterior abdominal wall. When the gallbladder has an unusually long mesentery, it can “wander” or “float” (wandering or free-floating gallbladder, WG), increasing the risk for torsion, as it can twist around its pedicle, leading to subsequent necrosis. In the literature, over 500 cases of gallbladder torsion, but less than 10 reports of WG [7] have been reported at the time of writing.

We describe a case where we detected intraoperatively a WG, which wrapped around the hepatoduodenal ligament. The patient was brought to the operating theatre with working diagnosis of a gastrointestinal stromal tumor (GIST), supported by CT scan.

CASE REPORT

A 77-year-old male with a medical history of hypertension, carrier of an aortic prosthesis for thoracic aneurysm, treated with endovascular techniques, with prior excision of *Kaposi's* sarcoma of the glans and of the leg, was admitted as an emergency case for abdominal pain, fever, and obstructive jaundice (hyperbilirubinemia, 3.30 mg/dL), with no biochemical signs of acute pancreatitis. An abdominal ultrasound showed a distended gallbladder with a large gallstone in the fundus, and a marked dilation of the intra and extra-hepatic biliary tract. A subsequent CT-scan confirmed the gallstone disease, and the distention of the proximal segment of the biliary tract, evidencing a normal size of the distal choledochus. Furthermore, CT-imaging (Figure 1) showed a thickening in the context of the submucosa of the anterior wall of the distal stomach that suggested a GIST of the gastric antrum. The working diagnosis was cholangitis in a patient with suspected GIST of the stomach. After initiating antibiotic therapy and restoring the hydro-electrolyte balance, the patient underwent an Endoscopic Retrograde Cholangio Pancreatography (ERCP). ERCP showed absence of gallstones in the common bile duct, the mid segment of which appeared thinned, while there was proximal dilation before the biliary *carrefour*. Moreover, a swelling of the gastric antrum wall in the presence of a normal mucosa was confirmed. A cholangio-

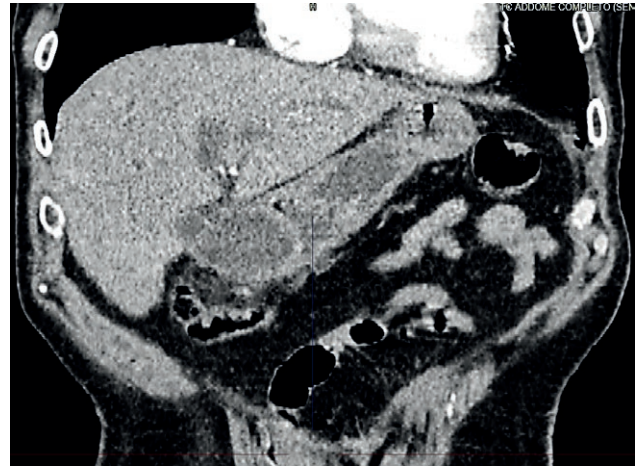


Figure 1. Coronal CT scan of the abdomen showing a tumor at the distal stomach with hypodense core, which was interpreted as suspect GIST.

MRI could not be performed due to the presence of a non-titanium prosthesis of the hip, and an endoscopic US was not exhaustive regarding the gastric wall. Based on this diagnostic picture, the patient was referred to laparoscopic surgery for a cholecystectomy and resection of the stomach. Intraoperatively, anatomical subversion of the standard anatomy was evident by a marked pattern of abdominal adhesions with diffuse post-inflam-



Figure 2. Schematic drawing depicting the gallbladder free from attachments to the liver and wrapped around the hepatic pedicle. The fundus of the gallbladder rests on the posterior portion of the gastric antrum.

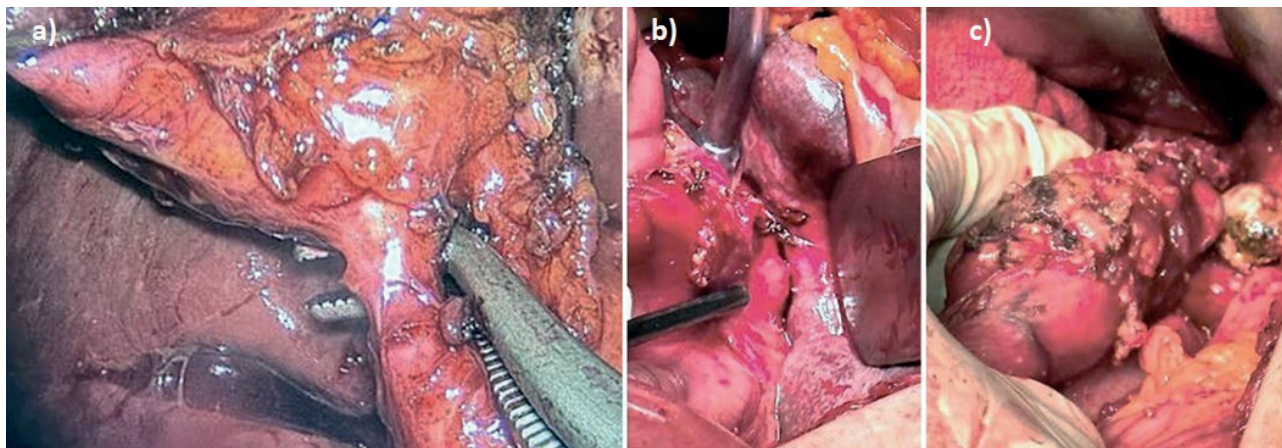


Figure 3. Intraoperative picture showing: a) laparoscopic view of the hepatic pedicle; b) adhesion of the gallbladder fundus to the gastric antrum in a fashion that suggested a fistulation process; c) gallbladder mobilized from the gastric fundus and repositioned to its original site.

matory scar tissue, resulting in the complete obfuscation of the hepatoduodenal ligament and the hepatic hilum (Figure 2). An intraoperative, laparoscopic US-scan was performed, documenting an area of fusion of the walls of the gallbladder and of the gastric antrum, with the interposition of a fluid-containing area of 2.5 cm of maximum diameter. This intraoperative picture suggested a cholecysto-gastric fistulation process, contraindicating further laparoscopy, so the procedure was converted to laparotomy (Figure 3).

Therefore, following the opening of the gastro-colic and gastro-hepatic ligament, gentle adhesiolysis, from the stomach and duodenum to the hepatic hilum, evidenced a wandering bulky gallbladder, wrapping anteriorly the hepatoduodenal ligament and the hepatic hilum. The fundus of the gallbladder containing a large stone appeared fused with the prepyloric antrum, forming a gallbladder-gastric fistula, mimicking the gastric swelling already demonstrated at preoperative imaging. Surgery was completed by standard cholecystectomy after repositioning the gallbladder to its natural location to the right of the common bile duct. An intra-operative trans-cystic duct cholangiogram showed normal anatomy of the biliary tract. Postoperative course was uneventful and the patient was discharged four days after surgery with normal serum bilirubin. Pathology confirmed inflammatory changes of the portion of gallbladder wall adherent to the stomach, representing the early stage of a cholecysto-gastric fistula.

DISCUSSION

Cholecystectomy for gallstone disease represents one of the most common procedures in routine surgical

practice, with a risk for post-operative mortality that has been estimated to be between 0.1 and 0.7% [8]. However, mortality increases to 3.6% when cholecystectomy is performed for acute cholecystitis[9, 10]. Mortality rates were not substantially affected by the introduction of a LC; LC is a widely accepted procedure, because postoperative recovery is rapid, it is more acceptable to patients and, consequently, it has replaced open cholecystectomy as the procedure of choice for the treatment of symptomatic gallbladder disease[10]. On the other hand, LC is the most frequent cause of injury to the bile duct, and more rarely to the hepatic artery and portal vein, duodenum, small bowel, and colon. These injuries tend to occur when the operation is difficult as a result of inflammation or subversion of anatomy, or resulting from trocar punctures, thermal injury, and difficult adhesiolysis[10-12].

Congenital anomalies of the gallbladder, its duct, and its blood supply, are sufficiently common for the surgeons that they need to be aware of these vagaries. Indeed, most postoperative complications of cholecystectomy may be related to a combination of inadequate anatomical knowledge or poor visualization of the operative field [13]. This determines a high rate of perioperative complications, but also of mortality. So, the results can be additional surgeries, long recovery time, loss of time from work, and reduction in the quality of life [14] [15] [16].

In WG there is no adhesion or even a loose attachment to the liver, so the gallbladder is only connected to the cystic duct and its mesentery. The first report of free-floating or wandering gallbladder was by Wendel in 1898 with a torsion of the gallbladder complicated by perforation [17]. Gallbladder torsion is largely acquired and secondary to aging but it may also be congenital. Generally, there is an abnormal migration of the *pars cystica*

Table 1. Wandering gallbladder.

N.	Author	Year of publication	Publication type	Age	Presenting signs/symptoms	Hospital admission
1	Wendel A.V.	1898	Case report	33	Abdominal pain	Emergency
2	Ziegler H.	1952	Case report	-	-	-
3	Maki et al	1962	Case report	-	-	-
4	Chiavarini RL et al	1975	Case report	22	Abdominal pain	Elective
5	Faso F.J. et al	1989	Case report	81	-	Emergency
6	Mathonnet et al	2001	Case report	-	-	-
7	Marano A. et al	2002	Case report	90	Abdominal pain, nausea, vomiting	Emergency
8	Otani et al	2007	Case report	57	Abdominal pain, pyrexia	Emergency
9	Morales A.M. et al	2008	Case report	70	Acute diarrhoea, back pain	Emergency
10	Boer J.	2011	Case report	89	Abdominal pain, nausea	Emergency
11	Warfe S. R. et al	2013	Case report	69	Acute epigastric pain, nausea	Emergency
12	Wu W-C et al	2013	Case report	40	Abdominal pain	Emergency
13	Kopp et al	2019	Case report	51	Hypertension, GE reflux, abdominal pain	Emergency
14	Miroslav K. et al	2019	Case report	51	Abdominal pain	Emergency

from the hepatic diverticulum during the fourth to seventh weeks of embryologic development. Aside an early description in 1952, a handful of articles on WG have since appeared (Table 1).

The clinical presentation varies among individuals. It can be asymptomatic, it may cause recurrent episodes of abdominal pain and transient hyperbilirubinemia, or present acutely with potential fatal torsion and necrosis due to its predisposition to twist around its vascular pedicle leading to ischemic necrosis[18]. The incidence of torsion is higher in the elderly population, with male to female ratio of 1:3 [19].

Preoperative diagnosis is difficult and rarely made by ultrasonography, and also CT. However, there are no specific radiologic signs, but a gallbladder with a long cystic duct that is not in its normal anatomic position can be suggestive [7]. A delay in the diagnosis may end up in rupture and peritonitis.

When a complete torsion occurs, a scenario of acute abdomen could appear. The mechanisms leading to torsion are poorly understood [7]. Three anatomic variants are believed to be responsible for torsion. The first variant is a free-floating gallbladder, suspended only by the cystic mesentery, where the gallbladder may rotate around its vascular axis represented by the cystic artery. Another variant is when the gallbladder hangs from the liver through an elongated, narrow-based and freely mobile mesentery, possibly due to age-related viscerop-tosis. Here rotation may occur around an axis represented by the gallbladder itself or its liver mesentery. A third variant described is extremely rare and consists of a normally fixed gallbladder to a mobile liver lobe free of its coronary and triangular ligaments[13]. Various trig-

gering mechanisms have been mentioned including visceroptosis, weight loss, gastric or colonic hyperperistalsis, constipation or diarrhoea, sigmoid volvulus, cholelithiasis, kyphosis, or iatrogenic manipulation. Patients with wandering gallbladder can present biliary colic-like symptoms attributed to torsion and de-torsion around the cystic duct as well as acute cholecystitis secondary to persistent obstruction[17-19]. In this reported case the encirclement of the hepatoduodenal ligament and adhesion to the stomach was evidently the result of a long and chronic process, with no acute signs of ischaemia.

A further peculiarity of this case was the misunderstanding of the gastric wall at imaging. So that the inflammatory reaction of the gallbladder wall with initial involvement of the adjacent stomach mimicked a swelling of the gastric wall, looking like a GIST.

Accurate knowledge of anatomical variants of the biliary anatomy is important when elaborating differential diagnosis for suspect gallbladder pathology and even more so when operating complex cases with a working diagnosis that may be misleading. In fact, a routine cholecystectomy may harbour unexpected and at times severe complications that may be anticipated and prevented when a complex picture can be reverted to normal anatomy.

CONCLUSION

A case is reported of wandering gallbladder wrapped around the hepatoduodenal ligament and adherent to the gastric fundus. Anomalies of position of the gallbladder are rare and should be suspected when gallblad-

der is not found in its classical position. Reverting to normal anatomy by careful dissection is the key to recognize anatomical variants and to prevent complications at surgery.

REFERENCES:

1. Lammert F, Gurusamy K, Ko CW, et al., Gallstones. 2016. **2**(1): p. 1-17.
2. Pisano M, Allievi N, Gurusamy K, et al., 2020 World Society of Emergency Surgery updated guidelines for the diagnosis and treatment of acute calculus cholecystitis. 2020. **15**(1): p. 1-26.
3. Törnqvist B, Strömberg C, Persson G, et al., Effect of intended intraoperative cholangiography and early detection of bile duct injury on survival after cholecystectomy: population based cohort study. 2012. **345**.
4. Fletcher DR, Hobbs MS, Tan P, et al., Complications of cholecystectomy: risks of the laparoscopic approach and protective effects of operative cholangiography: a population-based study. 1999. **229**(4): p. 449.
5. Sgaramella LI, Gurrado A, Pasculli A, et al., The critical view of safety during laparoscopic cholecystectomy: Strasberg Yes or No? An Italian Multicentre study. 2021. **35**: p. 3698-3708.
6. Gui D, Magalini S, Prete F, et al., What's right when the gallbladder's left? *Surgical Endoscopy*, 2002. **16**(11): p. 1637-1637.
7. Kopp M, Cornish NA, and Nazir SJIS, Wandering Gallbladder: A Case Report. 2019. **104**(7-8): p. 371-374.
8. Sandblom G, Videhult P, Crona Guterstam Y, et al., Mortality after a cholecystectomy: a population-based study. 2015. **17**(3): p. 239-243.
9. González-Castillo AM, Sancho-Insenser J, Miguel-Palacio D, et al., Mortality risk estimation in acute calculous cholecystitis: beyond the Tokyo Guidelines. 2021. **16**(1): p. 1-10.
10. Kwon A-H, Inui H, and Kamiyama YJWjos, Laparoscopic management of bile duct and bowel injury during laparoscopic cholecystectomy. 2001. **25**: p. 856-861.
11. El-Banna M, Abdel-Atty M, El-Meteini M, et al., Management of laparoscopic-related bowel injuries. 2000. **14**: p. 779-782.
12. Ress AM, Sarr MG, Nagorney DM, et al., Spectrum and management of major complications of laparoscopic cholecystectomy. 1993. **165**(6): p. 655-662.
13. Berger HJBMJ, Wandering Gall Bladders. 1975: p. 193.
14. Törnqvist B, Waage A, Zheng Z, et al., Severity of acute cholecystitis and risk of iatrogenic bile duct injury during cholecystectomy, a population-based case-control study. 2016. **40**: p. 1060-1067.
15. Melton GB, Lillemoe KD, Cameron JL, et al., Major bile duct injuries associated with laparoscopic cholecystectomy: effect of surgical repair on quality of life. 2002. **235**(6): p. 888.
16. Testini M, Piccinni G, Lissidini G, et al., Management of descending duodenal injuries secondary to laparoscopic cholecystectomy. 2008. **25**(1): p. 12-15.
17. AV W, A case of floating gallbladder and kidney complicated by cholelithiasis with perforation of the gallbladder. *Ann Surg*, 1898. **27**: p. 199-202.
18. Morales AM and Tyroch AHJTAjos, Wandering gallbladder. 2008. **196**(2): p. 240-241.
19. Quinn SF, Fazzio F, and Jones EJAjor, Torsion of the gallbladder: findings on CT and sonography and role of percutaneous cholecystostomy. 1987. **148**(5): p. 881-882.