Case Report

Gallbladder Volvulus in a Patient with Type I Choledochal Cyst: A Case Report and Review of the Literature

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1. Introduction

Gallbladder volvulus is a rare, difficult-to-diagnose clinical condition that presents as acute abdomen and often leads to significant morbidity due to the delay in diagnosis and surgical treatment. Since first reported by Wendel in 1898 as a “floating gallbladder,” more than five hundred cases have been reported in the literature with an incidence of approximately 1 in 365,000 hospital admissions [1, 2]. Volvulus of the gallbladder usually presents in elderly frail women in their seventh or eighth decade of life [3]. With prompt surgical intervention, the potential for gallbladder gangrene and perforation can be averted and an excellent prognosis is achieved [4–6]. Choledochal cysts are also uncommon pathological dilations of the biliary tree, first reported in [7]. While being associated with multiple congenital anomalies of the hepatobiliary and pancreatic systems, they have not been reported to increase the likelihood of gallbladder volvulus. Herein we describe a case of successful surgical treatment of gallbladder volvulus in an elderly woman with concomitant Todani type IC choledochal cyst, shedding the light on the perioperative management of an extremely rare biliary tree disease combination [8].

2. Case Presentation

A 92-year-old woman presented to the emergency room with acute onset of right upper quadrant pain, nausea, and vomiting. She was previously in her usual state of health with minimal medical or surgical comorbidities with the exception of a minor weight loss. Her past medical history is significant for gastroesophageal reflux, history of peptic ulcer disease, and osteoarthritis. She has had cataract surgery in the past in addition to colonic polypectomy for benign polyps a few years prior to her presentation. Her family history was significant for uterine cancer in her mother, without known genetic predisposition. On physical examination, she was hemodynamically normal and the only pertinent finding was tenderness to palpation in her right upper and lower abdominal quadrants. All blood tests were within normal limits including a white blood cell count and liver function...
tests. A right upper quadrant ultrasound demonstrated gallbladder distention with mild gallbladder wall thickening, but no evidence of gallstones, sludge, or pericholecystic fluid. A computed tomography (CT) scan demonstrated a markedly dilated extrahepatic biliary ductal system (intrapancreatic common bile duct > 2.5 cm) and a markedly distended gallbladder (Figure 1). The patient was admitted to the hospital for treatment of acute cholecystitis including bowel rest and antibiotics. Given the marked dilation of the intra- and extrahepatic bile ducts an obstructing mass at the head of the pancreas or the distal bile duct had to be ruled out. Magnetic resonance cholangiopancreatography (MRCP) demonstrated a distended gallbladder with wall thickening, edema, and pericholecystic fluid confirming acalculous cholecystitis, with a mention of focal narrowing and wall thickening of the cystic duct. The extrahepatic bile duct had a fusiform dilation measuring 27 mm in the largest dimension (Figure 2). Endoscopic retrograde cholangiopancreatography (ERCP) and an endoscopic ultrasound (EUS) failed to show a pancreatic head mass or distal common bile duct stricture; however, the cystic duct did not opacify, suggesting an obstruction in the absence of gallstones. The patient’s clinical status did not improve during a short period of observation; she developed increasing abdominal pain and leukocytosis to \(13.5 \times 10^3 / \mu L\). After reviewing of previous CT scans spanning several years, the gallbladder was noted to be located in several different locations adding suspicion for gallbladder torsion to the differential diagnosis (Figure 1). The patient was consented for open cholecystectomy due to the extremely large size of her gallbladder, her concomitant small body habitus, and a relative delay in diagnosis.

At laparotomy, a gangrenous, necrotic gallbladder was identified in the right lower quadrant and was nonadherent to the liver bed. The gallbladder was completely torse (360 degrees) around the cystic duct and cystic artery. The gallbladder was so mobile that it could be brought out onto the abdominal wall and detorsed; a blood clot was visible in the gallbladder mesentery at the point of torsion. The gallbladder was readily removed following simple ligation of the cystic artery and duct (Figure 3). The choledochal cyst was not addressed given her advanced age and lack of malignancy seen on axial imaging, ERCP, and EUS. Inspection of the specimen revealed significant gallbladder wall thickening and absence of gallstones (Figure 3). The patient recovered without major events and was discharged to a rehabilitation facility on postoperative day four. Final pathology revealed severe acute cholecystitis with transmural necrosis and acute serositis.
3. Discussion

Gallbladder volvulus or torsion occurs when the gallbladder rotates either clockwise or counterclockwise around its mesentery along the axis of the cystic duct and cystic artery causing complete obstruction of blood flow and biliary drainage resulting in acute gangrenous cholecystitis [3]. This condition is extremely rare; it occurs in less than 0.1% patients who undergo urgent cholecystectomies for presumed acute cholecystitis [4]. The incidence of gallbladder volvulus increases with age and peaks in subjects between sixty and eighty years of age. It is more common in women, with a female: male ratio of 3-4:1 [2].

The etiology of gallbladder torsion is unknown. Anatomic anomalies, such as an abnormally long mesentery or abnormal fixation of the gallbladder to the liver, can result in a suspended gallbladder allowing it to freely float from the liver bed [3]. It is thought to occur more commonly in the elderly due to the loss of visceral fat, liver atrophy, and increased elasticity that allows the gallbladder to freely hang or “float.” Significant peristaltic movements of the stomach, duodenum, and nearby colon, in addition to mechanical predisposition caused by kyphoscoliosis, have been cited in the literature as predisposing factors for gallbladder torsion [9, 10]. Gallstones are considered incidental rather than causative when found in patients with gallbladder volvulus. They are only present...
in 25–50% of patients with gallbladder torsion, and thus they are unlikely to be the underlying etiology [10].

Patients with gallbladder torsion present with symptoms similar to biliary colic or acute cholecystitis including acute onset of right upper quadrant pain. The acuteness of the presentation depends on whether the volvulus is partial or complete; a partial torsion is followed by spontaneous detorsion and resolution of symptoms, thus presenting like bile spillage and peritonitis. While laparoscopic detorsion and cholecystectomy is effective in the management of gallbladder torsion, we chose open cholecystectomy due to the very large size of the gallbladder reaching a pelvic location in a patient with an extremely small body habitus (no room for laparoscopic instrumentation/visualization) [23, 24]. Laparoscopic cholecystectomy and dissection of the critical view of safety in gallbladder torsion patients is usually straightforward given the lack of a cystic plate attaching the gallbladder to the liver [12, 23].

To our knowledge, this is the first report in the literature that describes the presence of gallbladder torsion in a patient with a choledochal cyst. Choledochal cysts have been associated with other anomalies within the pancreaticobiliary tree including multiseptated gallbladder, heterotopic pancreas, and pancreatic divisum, but they have not been reported to predispose to volvulus of the gallbladder [8, 25]. Choledochal cysts are classified into five types based on the Alonso-Lej modification of the initial Todani classification [26]. Our patient had a type I choledochal cyst which is the most common cyst type [7]. The presence of a choledochal cyst is considered an indication for surgical resection as many of these become symptomatic, presenting with recurrent cholangitis or pancreatitis and harbor a risk of synchronous cholangiocarcinoma [27, 28]. In a large, multi-institutional western study that included 394 patients, cyst excision to include extrahepatic bile duct resection and hepaticoenterostomy was the standard of surgical care and completed in 80.3% of patients [7]. Malignancy was found in 9.1% and at follow-up, 3.3% of patients had cancer recurrence with a median survival of 3.5 years [7]. This study reconfirmed prophylactic surgical resection of choledochal cysts.

Due to the rarity of this combination of gallbladder torsion and choledochal cyst, a hepatobiliary surgeon should be involved from the outset. Prompt gallbladder detorsion and cholecystectomy is the standard of care for gallbladder volvulus regardless of the timing of diagnosis (pre- or intraoperatively). Extrapolation from common surgical practice precludes excision of the choledochal cyst and construction of a hepaticoenterostomy in the setting of an acutely inflamed and potentially infected field; thus laparoscopic cholecystectomy should be done first. The patient can then be brought back electively for choledochal cyst resection. In cases of acute cholecystitis without gallbladder torsion, conservative management of cholecystitis can be attempted with antibiotics with or without percutaneous cholecystostomy drainage which may then allow for a one-stage procedure (cholecystectomy and choledochal cyst resection). We suggest an algorithm extrapolated from literature data suggesting a plan of action when a gallbladder volvulus is encountered in a patient with choledochal cysts (Figure 4). It is based on the patient’s clinical status and the chronicity of the volvulus in addition to the cyst type, as the procedures needed to treat choledochal cysts vary from a simple ERCP/sphincterotomy...
4. Conclusion

Gallbladder volvulus and choledochal cysts are two very rare entities of the biliary tree. Volvulus should be considered in the elderly female patients with symptoms suggestive of sudden onset of acute acalculous cholecystitis with imaging characteristics pointing towards a free-floating gallbladder. While gallbladder volvulus requires a high index of suspicion and early surgical intervention to avoid significant morbidity and mortality in a frail patient population, choledochal cysts can be dealt with electively taking into account their symptoms and risk for malignancy relative to age.

Competing Interests

The authors declare that they have no competing interests.

References

[1] A. V. Wendel, “VI. A case of floating gall-bladder and kidney complicated by cholelithiasis, with perforation of the gall-bladder,” *Annals of Surgery*, vol. 27, no. 2, pp. 199–202, 1898.

[2] D. J. Reilly, G. Kalogeropoulos, and D. Thiruchelvam, “Torsion of the gallbladder: a systematic review,” *HPB*, vol. 14, no. 10, pp. 669–672, 2012.

[3] N. J. Mouawad, B. Crofts, R. Streu, R. Desrochers, and B. C. Kimball, “Acute gallbladder torsion—a continued pre-operative diagnostic dilemma,” *World Journal of Emergency Surgery*, vol. 6, no. 1, article 13, 2011.

[4] A. Nakao, T. Matsuda, S. Funabiki et al., “Gallbladder torsion: case report and review of 245 cases reported in the Japanese literature,” *Journal of Hepato-Biliary-Pancreatic Surgery*, vol. 6, no. 4, pp. 418–421, 1999.

[5] S. Y. Kim and J. T. Moore, “Volvulus of the gallbladder: laparoscopic detorsion and removal,” *Surgical Endoscopy*, vol. 17, no. 11, p. 1849, 2003.

[6] H. A. Amarillo, E. D. Pirchi, and M. E. Mihura, “Complete gallbladder and cystic pedicle torsion. Laparoscopic diagnosis and treatment,” *Surgical Endoscopy*, vol. 17, no. 5, pp. 832–833, 2003.

[7] K. C. Soares, Y. Kim, G. Spolverato et al., “Presentation and clinical outcomes of choledochal cysts in children and adults: a multi-institutional analysis,” *JAMA Surgery*, vol. 150, no. 6, pp. 577–584, 2015.

[8] T. Todani, Y. Watanabe, M. Narusue, K. Tabuchi, and K. Okajima, “Congenital bile duct cysts: classification, operative procedures, and review of thirty-seven cases including cancer arising from choledochal cyst,” *The American Journal of Surgery*, vol. 134, no. 2, pp. 263–269, 1977.

[9] P. C. Garciavilla, J. F. Alvarez, and G. V. Uzqueda, “Diagnosis and laparoscopic approach to gallbladder torsion and cholelithiasis,” *Journal of the Society of Laparoendoscopic Surgeons*, vol. 14, no. 1, pp. 147–151, 2010.

[10] A. C. Stieber and J. J. Bauer, “Volvulus of the gallbladder,” *The American Journal of Gastroenterology*, vol. 78, no. 2, pp. 96–98, 1983.

[11] A. A. Shaikh, A. Charles, S. Domingo, and G. Schaub, “Gallbladder volvulus: report of two original cases and review of the literature,” *The American Surgeon*, vol. 71, no. 1, pp. 87–89, 2005.

[12] E. A. Boonstra, B. van Etten, T. R. Prins, E. Sieders, and B. L. van Leeuwen, “Torsion of the gallbladder,” *Journal of Gastrointestinal Surgery*, vol. 16, no. 4, pp. 882–884, 2012.

[13] M. S. Vedanayagam, I. Nikolopoulos, G. Janakan, and A. El-Gaddal, “Gallbladder volvulus: a case of mimicry,” *BMJ Case Reports*, vol. 2013, 2013.

[14] G. Janakan, A. A. Ayantunde, and H. Hoque, “Acute gallbladder torsion: an unexpected intraoperative finding,” *World Journal of Emergency Surgery*, vol. 3, no. 1, article 9, 2008.

[15] G. C. Christoudias, “Gallbladder volvulus with gangrene. Case report and review of the literature,” *Journal of the Society of Laparoendoscopic Surgeons*, vol. 1, no. 2, pp. 167–170, 1997.
[16] E. Hinoshita, T. Nishizaki, K. Wakasugi et al., “Pre-operative imaging can diagnose torsion of the gallbladder: report of a case,” *Hepato-Gastroenterology*, vol. 46, no. 28, pp. 2212–2215, 1999.

[17] Y. Tajima, N. Tsunekawa, T. Kuroki, and T. Kanematsu, “Gallbladder torsion showing a ‘whirl sign’ on a multidetector computed tomography scan,” *American Journal of Surgery*, vol. 197, no. 1, pp. e9–e10, 2009.

[18] G. J. Wang, M. Collin, J. Crossett, and R. A. Holmes, “Bulls-eye image of gallbladder volvulus,” *Clinical Nuclear Medicine*, vol. 12, no. 3, pp. 231–232, 1987.

[19] M. Fukuchi, K. Nakazato, H. Shoji, H. Naitoh, and H. Kuwano, “Torsion of the gallbladder diagnosed by magnetic resonance cholangiopancreatography,” *International Surgery*, vol. 97, no. 3, pp. 235–238, 2012.

[20] M. Usui, S. Matsuda, H. Suzuki, and Y. Ogura, “Preoperative diagnosis of gallbladder torsion by magnetic resonance cholangiopancreatography,” *Scandinavian Journal of Gastroenterology*, vol. 35, no. 2, pp. 218–222, 2000.

[21] T. Koyanagi and K. Sato, “Complete gallbladder torsion diagnosed with sequential computed tomography scans: a case report,” *Journal of Medical Case Reports*, vol. 6, article 289, 2012.

[22] B. J. Potter, L. Alfar, and H. W. Hollis, “A clinician’s guide to the diagnosis and management of gallbladder volvulus,” *The Permanente Journal*, vol. 17, no. 2, pp. 80–83, 2013.

[23] Y. K. Sunder, S. P. Akhilesh, G. Raman, S. Deborshi, and M. H. Shantilal, “Laparoscopic management of a two staged gall bladder torsion,” *World Journal of Gastrointestinal Surgery*, vol. 7, no. 12, pp. 403–407, 2015.

[24] T. Nguyen, A. Geraci, and J. J. Bauer, “Laparoscopic cholecystectomy for gallbladder volvulus,” *Surgical Endoscopy*, vol. 9, no. 5, pp. 519–521, 1995.

[25] K. C. Soares, D. J. Arnaoutakis, I. Kamel et al., “Choledochal cysts: presentation, clinical differentiation, and management,” *Journal of the American College of Surgeons*, vol. 219, no. 6, pp. 1167–1180, 2014.

[26] F. Alonso-Lej, W. B. Rever Jr., and D. J. Pessagno, “Congenital choledochal cyst, with a report of 2, and an analysis of 94, cases,” *International Abstracts of Surgery*, vol. 108, no. 1, pp. 1–30, 1959.

[27] P. A. Lipsett, H. A. Pitt, P. M. Colombani, J. K. Boitnott, and J. L. Cameron, “Choledochal cyst disease: a changing pattern of presentation,” *Annals of Surgery*, vol. 220, no. 5, pp. 644–652, 1994.

[28] B. H. Edil, K. Olino, and J. L. Cameron, “The current management of choledochal cysts,” *Advances in Surgery*, vol. 43, no. 1, pp. 221–232, 2009.