Case Report

Spontaneous retroperitoneal bilious collection: A case report

Golnaz Moradi, MD a, Mohaddeseh Rezaei b and Seyed A. Miratashi Yazdi, MD c, *

a Department of Radiology, Sina Hospital, Tehran University of Medical Sciences, Tehran, Iran
b Students' Scientific Research Center, Tehran University of Medical Sciences, Tehran, Iran
c Department of Surgery, Sina Hospital, Tehran University of Medical Sciences, Tehran, Iran

Received 15 August 2021; revised 26 October 2021; accepted 4 November 2021; Available online 9 December 2021

Abstract

Biloma refers to the encapsulated accumulation of bile in the abdomen. Bilomas generally occur after bile duct disruption. The clinical case of the present report was a 75 Y/O woman with jaundice, fever, abdominal pain, lethargy, and anorexia who had been admitted to the emergency department. Endoscopic retrograde cholangiopancreatography (ERCP) showed severe diffuse dilatation of the common bile duct (CBD) and common hepatic duct (CHD) with filling defects. One stone and some sludge and pus were extracted after balloon swiping. Plastic CBD stent placement was performed. An abdominal ultrasound scan showed a stone in the CBD and fluid collection in the right perirenal space. Under ultrasound guidance, a drainage tube was inserted into the retroperitoneal biloma. A serial ultrasound scan revealed a gradual decrease in the size of the biloma.

Perforation of the bile duct, which is characterised by the collection of retroperitoneal fluid, is considered an extremely rare condition that can be fatal. Ultrasonography, computed tomography, and endoscopic retrograde cholangiopancreatography were used for the diagnosis. The treatment includes prevention of leakage and bile drainage.

Keywords: Bile accumulation; Biloma; Cholangiopancreatography; Endoscopic retrograde; Hepatic duct, Common; Retroperitoneum

© 2021 The Authors.
Production and hosting by Elsevier Ltd on behalf of Taibah University. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/). https://doi.org/10.1016/j.jtumed.2021.11.001

Introduction

Biloma refers to the encapsulated accumulation of bile in the abdomen. Bilomas generally occur after bile duct disruption. The existing literature shows that the main causes
of this problem include iatrogenic (following hepatobiliary surgery), trauma, or problems due to choledocholithiasis.

Retroperitoneal accumulation of bile and non-traumatic rupture of the biliary tract, which is spontaneous, is an extremely special condition. Impulsive perforation of the bile duct is not common among adults, particularly in cases that accompany retroperitoneal biloma. In this study, a patient with retroperitoneal biloma due to cholangitis and choledocholithiasis is reported.

Case presentation

A 75 years old woman was admitted to the emergency department with abdominal pain that had begun 72 h before admission. Along with pain, jaundice, fever, lethargy, and anorexia were observed. The patient’s medical history showed Hypertension and open cholecystectomy 15 years prior. Examination of the patient revealed distention of the patient’s abdomen. In addition, the right upper quadrant of the abdomen and the epigastric region were tender. In the abdomen, no rebound tenderness or guarding was observed. The patient had a fever of 38.9 °C.

Lab test results are presented in Table 1. The rest of the laboratory test results were normal.

![Image of ultrasound scan](image1.png)

Figure 1: Fluid collection in right perirenal space and dilated CBD containing a stone.

An ultrasound scan of the abdomen revealed fluid accumulation in the right perirenal space, dilatation of intrahepatic and extrahepatic bile ducts, and sludge and a stone in the CBD (Figure 1).

Computed tomography revealed fluid collection in the anterior and posterior pararenal spaces on the right side, as well as dilatation of the biliary tract (Figure 2).

Endoscopic retrograde cholangiopancreatography (ERCP) revealed severe diffuse dilation of the common hepatic duct (CHD) and common bile duct (CBD) containing filling defects. A sphincterotomy was performed. After balloon sweeping, one stone was extracted, and some sludge and pus were extracted. Plastic CBD stent placement was performed.

In percutaneous aspiration of the retroperitoneal fluid, which was carried out by ultrasound, a biliary fluid was found to be negative for bacteria. Regarding the diagnosis of retroperitoneal biloma, re-ERCP was performed, which revealed diffuse dilatation of the bile ducts. The previous CBD stent was removed. No obvious leakage was observed. Because of the symptoms of cholangitis, a plastic stent was placed. A drainage tube was inserted into the retroperitoneal biloma under ultrasound guidance. The drainage output was biliary coloured. In serial ultrasound scans, the size of the biloma decreased gradually. Despite the removal of the drainage tube, a follow-up ultrasound scan after one month showed no fluid collection in the abdomen.

Discussion

A biloma refers to an accumulation of bile that occurs in the exterior parts of the biliary system and in the abdominal area and may be extrahepatic or intrahepatic. The most prevalent causes of biloma are hepatobiliary interventions and laparoscopic cholecystectomy, after which choledocholithiasis, abdominal trauma, and biliary dilatation following biliary stricture are observed. Most bilomas are iatrogenic or traumatic. Spontaneous biloma is not common, and most of them are observed with choledocholithiasis or cholangiocarcinoma.

The major perforation site of the biliary tract is the GB, and the cause is usually cholecystitis and cholecystolithiasis. The CBD was another perforation site. Bile duct perforation

| Lab tests                  | Results | References | Unit      |
|---------------------------|---------|------------|-----------|
| Leukocytes                | 15      | 4–10.5     | 10^3/μl   |
| Neutrophils               | 90.8    | 54–62      | %         |
| Haemoglobin               | 12.1    | 12.5–16    | g/dl      |
| Platelets                 | 172     | 150–400    | 10^3/μl   |
| Albumin                   | 2.9     | 3.5–5.2    | g/dl      |
| Amylase                   | 38      | Up to 90   | IU/L      |
| Total bilirubin           | 3.59    | 0.3–1.2    | mg/dl     |
| Direct bilirubin          | 2.35    | Up to 0.4  | mg/dl     |
| Indirect bilirubin        | 1.24    | 0.2–0.8    | mg/dl     |
| Alkaline phosphatase      | 249     | Up to 240  | IU/l      |
| Aspartate aminotransferase| 13      | Up to 31   | IU/l      |
| Alanine aminotransferase  | 64      | Up to 31   | IU/l      |
| Prothrombin time          | 15.9    | 12–14      | sec       |
| Partial thromoplastin time| 44.4    | 25–38      | sec       |
| C-reactive protein        | 61.4    | 0–10; Neg  | mg/L      |

![Table 1: Lab tests result on admission.](image2.png)
is shown as either localised fluid accumulation or general biliary peritonitis. The reason for bile leakage may be the retroperitoneal portion. One pathophysiological cause is a perforation in the gallbladder. This adheres to the parietal peritoneum through a chronic inflammatory process. When this happens, communication exists between the retroperitoneum and the biliary tract.

As it is known, the most common cause of spontaneous biloma is choledocholithiasis. On the other hand, the secondary cause of spontaneous bile duct perforation may be due to the erosion of gallstones. Increased intraductal pressure may also be due to obstruction of the distal bile duct (due to a stone, mass, or spasm of the odend sphincter), thrombosis of a vessel supplying the bile duct wall, cholangitis, regurgitation of pancreatic secretions into the bile duct, choledochocele, and acute pancreatitis.

In the present case, a stone existed in the CBD; however, no obvious bile leak was observed on ERCP. Ultrasound and computed tomography (CT) scan showed no choledochal cysts and no evidence of pancreatitis. Perforation of the biliary tract may be caused by choledocholithiasis or cholangitis.

In terms of incidence, there is no difference between females and males; however, the condition is usually observed in the age period of the 60s and 70s. The presentation is nonspecific, with abdominal pain, usually in the right upper quadrant. Fever may accompany abdominal distension and jaundice. In the present case, a 75 Y/O woman with pain in the right upper quadrant of the abdomen and fever with jaundice was admitted.

Blood tests may reveal an increase in CRP and leukocyte counts and liver function tests. In the present case, the CRP level was elevated to 61.4 mg/L and liver function tests were disrupted. Gram-negative bacteraemia may be revealed by blood cultures.

Biloma may be detected by magnetic resonance imaging, ultrasound, or CT scan. Abdominal ultrasound is the first imaging modality that leads to the suspicion of a cholero-peritoneum, because it identifies perirenal and retroperitoneal fluid accumulation, which may be misunderstood as a renal abscess. In most cases, dilatation of the bile ducts is observed, and retroperitoneal bile will be revealed by needle aspiration of the collection. In the present case, a diagnosis of biloma was made after percutaneous aspiration.

ERCP can help identify an active leak that can also be therapeutically intervened. Identifying the leakage site will also be difficult because, in most cases, it will have closed. Endoscopic treatment in cases where it is suitable includes sphincterotomy with stone extraction to lower biliary pressure. As another possible treatment, a stent can be placed in more distal lesions, since this results in a reduction in the pressure gradient into the duodenum and simplifies the flow of bile. This also releases obstruction from lesions that tighten the biliary tree.

In this case, due to the patient’s cholangitis, imipenem was prescribed empirically. Also, apart from external drainage, biliary stent was inserted. Non-surgical intervention before definitive biliary surgery is considered the first choice for treating biloma, as done in this case.

Conclusion

Perforation of the bile duct, which is characterised by the collection of retroperitoneal fluid, is considered an extremely rare condition, which can be fatal. Biloma should be considered in the differential diagnosis of retroperitoneal fluid collection without an air bubble.

The diagnosis and treatment of damages to the biliary tract are considered a challenge. Luckily, the process of diagnosis and treatment has been improved by new technologies, and many patients are treated without sequelae. In this case, after non-surgical interventions, the size of the biloma on serial ultrasound scans decreased gradually and disappeared eventually.
Source of funding

This research did not receive any grant from funding agencies in the public, commercial, or not-for-profit sectors.

Conflict of interest

The authors have no conflict of interest to declare.

Ethical approval

The patient was aware and content that her clinical data would be used for publication purposes and provided her consent.

Consent

Informed consent was obtained from the patient for this case report publication and any accompanying images.

Authors contributions

SAMY conceived and designed the analysis; SAMY and GM collected the data; MR and GM contributed data and wrote the paper. All authors have critically reviewed and approved the final draft and are responsible for the content and similarity index of the manuscript.

References

1. Trivedi PJ, Gupta P, Phillips-Hughes J, Ellis A. Biloma: an unusual complication in a patient with pancreatic cancer. World J Gastroenterol 2009 Nov 7; 15(41): 5218–5220.
2. Yousaf MN, D’Souza RG, Chaudhary F, Ehsan H, Sittambalam C. Biloma: a rare manifestation of spontaneous bile leak. Cureus 2020; 12(5).
3. Vazquez JL, Thorsen MK, Dodds WJ, Quiroz FA, Martinez ML, Lawson TL, et al. Evaluation and treatment of intraabdominal bilomas. Am J Roentgenol 1985; 144(5): 933–938.
4. Fujiwara H, Yamamoto M, Takahashi M, Ishida H, Ohashi O, Onoyama H, et al. Spontaneous rupture of an intrahepatic bile duct with biloma treated by percutaneous drainage and endoscopic sphincterotomy. Am J Gastroenterol 1998; 93(11): 2282–2284. Elsevier.
5. Roslyn J, Busuttil RW. Perforation of the gallbladder: a frequently mismanaged condition. Am J Surg 1979; 137(3): 307–312. Elsevier.
6. Ishii K, Matsuo K, Seki H, Yasui N, Sakata M, Shimada A, et al. Retroperitoneal biloma due to spontaneous perforation of the left hepatic duct. Am J Case Rep 2016; 17: 264. International Scientific Information, Inc.
7. Kang S-B, Han H-S, Min SK, Lee HK. Nontraumatic perforation of the bile duct in adults. Arch Surg 2004; 139(10): 1083–1087. American Medical Association-American Medical Association.
8. Kaushik R, Attri AK. Choloretroperitoneum—an unusual complication of cholelithiasis. Medknow Publications on behalf of Association of Surgeons of India; 2004.
9. Akhtar MA, Bandyopadhyay D, Montgomery HD, Mahomed A. Spontaneous idiopathic subcapsular biloma. J Hepatobiliary Pancreat Surg 2007; 14(6): 579–581. Epub 2007 Nov 30.
10. Mizutani S, Yagi A, Watanabe M, Maejima K, Komine O, Yoshino M, et al. Tube drainage for spontaneous perforation of the extrahaepatic bile duct. Med Sci Monit 2011 Jan; 17(1): CS8–11.
11. Garg PK, Jain BK, Pandey SD, Rathi V, Puri AS. Simultaneous non-traumatic perforation of the right hepatic duct and gallbladder. Malays J Med Sci 2012 Jul; 19(3): 77–80.
12. Faridi SH, Aslam M, Siddiqui B, Khan RA. Challenges in the diagnosis and management of spontaneous bile duct perforation: a case report and review of the literature. J Indian Assoc Pediatr Surg 2015; 20: 143–145.
13. Hartle RJ, McGarrity TJ, Conter RL. Treatment of a giant biloma and bile leak by ERCP stent placement. Am J Gastroenterol 1993; 88(12): 2117–2118.
14. Bas G, Okan I, Sahin M, Eryilmaz R, Isk A. Spontaneous biloma managed with endoscopic retrograde cholangiopancreatography and percutaneous drainage: a case report. J Med Case Rep 2011; 5(1): 1–3. Springer.
15. Lilly JR, Weintraub WH, Altman RP. Spontaneous perforation of the extrahaepatic bile ducts and bile peritonitis in infancy. Surgery 1974; 75(5): 664–673. Elsevier.
16. Takahashi K, Okabe Y, Orino A, Imai Y, Yazumi S, Chiba T. Spontaneous rupture of a biliary diverticulum in the distal common bile duct, with formation of a retroperitoneal biloma. Gastrointest Endosc 2005; 61(6): 783–787. Elsevier.

How to cite this article: Moradi G, Rezaei M, Miratashi Yazdi SA. Spontaneous retroperitoneal bilious collection: A case report. J Taibah Univ Med Sc 2022;17(4):619–622.