

World Journal of *Clinical Cases*

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REVIEW

- 2976 Gut microbiota in gastrointestinal diseases during pregnancy
Liu ZZ, Sun JH, Wang WJ
- 2990 Targeting metabolism: A potential strategy for hematological cancer therapy
Tang X, Chen F, Xie LC, Liu SX, Mai HR

MINIREVIEWS

- 3005 Elevated intra-abdominal pressure: A review of current knowledge
Łagosz P, Sokolski M, Biegus J, Tycinska A, Zymlinski R

ORIGINAL ARTICLE**Case Control Study**

- 3014 Changes in corneal nerve morphology and function in patients with dry eyes having type 2 diabetes
Fang W, Lin ZX, Yang HQ, Zhao L, Liu DC, Pan ZQ
- 3027 Combined sevoflurane-dexmedetomidine and nerve blockade on post-surgical serum oxidative stress biomarker levels in thyroid cancer patients
Du D, Qiao Q, Guan Z, Gao YF, Wang Q

Retrospective Cohort Study

- 3035 Early warning prevention and control strategies to reduce perioperative venous thromboembolism in patients with gastrointestinal cancer
Lu Y, Chen FY, Cai L, Huang CX, Shen XF, Cai LQ, Li XT, Fu YY, Wei J
- 3047 Dose-response relationship between risk factors and incidence of COVID-19 in 325 hospitalized patients: A multicenter retrospective cohort study
Zhao SC, Yu XQ, Lai XF, Duan R, Guo DL, Zhu Q

Retrospective Study

- 3060 Preventive online and offline health management intervention in polycystic ovary syndrome
Liu R, Li M, Wang P, Yu M, Wang Z, Zhang GZ
- 3069 Evidence-based intervention on postoperative fear, compliance, and self-efficacy in elderly patients with hip fracture
Fu Y, Zhu LJ, Li DC, Yan JL, Zhang HT, Xuan YH, Meng CL, Sun YH
- 3078 Significance of dysplasia in bile duct resection margin in patients with extrahepatic cholangiocarcinoma: A retrospective analysis
Choe JW, Kim HJ, Kim JS

- 3088** Diagnostic value and safety of medical thoracoscopy for pleural effusion of different causes

Liu XT, Dong XL, Zhang Y, Fang P, Shi HY, Ming ZJ

Observational Study

- 3101** Oxaliplatin-induced neuropathy and colo-rectal cancer patient's quality of life: Practical lessons from a prospective cross-sectional, real-world study

Prutianu I, Alexa-Stratulat T, Cristea EO, Nicolau A, Moisuc DC, Covrig AA, Ivanov K, Croitoru AE, Miron MI, Dinu MI, Ivanov AV, Marinca MV, Radu I, Gafton B

- 3113** Breast-conserving surgery and sentinel lymph node biopsy for breast cancer and their correlation with the expression of polyligand proteoglycan-1

Li FM, Xu DY, Xu Q, Yuan Y

SYSTEMATIC REVIEWS

- 3121** Clinical significance of aberrant left hepatic artery during gastrectomy: A systematic review

Tao W, Peng D, Cheng YX, Zhang W

META-ANALYSIS

- 3131** Betel quid chewing and oral potential malignant disorders and the impact of smoking and drinking: A meta-analysis

Lin HJ, Wang XL, Tian MY, Li XL, Tan HZ

- 3143** Effects of physical exercise on the quality-of-life of patients with haematological malignancies and thrombocytopenia: A systematic review and meta-analysis

Yang YP, Pan SJ, Qiu SL, Tung TH

CASE REPORT

- 3156** Primary malignant peritoneal mesothelioma mimicking tuberculous peritonitis: A case report

Lin LC, Kuan WY, Shiu BH, Wang YT, Chao WR, Wang CC

- 3164** Endoscopic submucosal dissection combined with adjuvant chemotherapy for early-stage neuroendocrine carcinoma of the esophagus: A case report

Tang N, Feng Z

- 3170** Lymph-node-first presentation of Kawasaki disease in a 12-year-old girl with cervical lymphadenitis caused by *Mycoplasma pneumoniae*: A case report

Kim N, Choi YJ, Na JY, Oh JW

- 3178** Tuberculosis-associated hemophagocytic lymphohistiocytosis misdiagnosed as systemic lupus erythematosus: A case report

Chen WT, Liu ZC, Li MS, Zhou Y, Liang SJ, Yang Y

- 3188** Migration of a Hem-o-Lok clip to the renal pelvis after laparoscopic partial nephrectomy: A case report

Sun J, Zhao LW, Wang XL, Huang JG, Fan Y

- 3194** Ectopic intrauterine device in the bladder causing cystolithiasis: A case report
Yu HT, Chen Y, Xie YP, Gan TB, Gou X
- 3200** Giant tumor resection under ultrasound-guided nerve block in a patient with severe asthma: A case report
Liu Q, Zhong Q, Zhou NN, Ye L
- 3206** Myomatous erythrocytosis syndrome: A case report
Shu XY, Chen N, Chen BY, Yang HX, Bi H
- 3213** Middle thyroid vein tumor thrombus in metastatic papillary thyroid microcarcinoma: A case report and review of literature
Gui Y, Wang JY, Wei XD
- 3222** Severe pneumonia and acute myocardial infarction complicated with pericarditis after percutaneous coronary intervention: A case report
Liu WC, Li SB, Zhang CF, Cui XH
- 3232** IgA nephropathy treatment with traditional Chinese medicine: A case report
Zhang YY, Chen YL, Yi L, Gao K
- 3241** Appendico-vesicocolonic fistula: A case report and review of literature
Yan H, Wu YC, Wang X, Liu YC, Zuo S, Wang PY
- 3251** *Scedosporium apiospermum* infection of the lumbar vertebrae: A case report
Shi XW, Li ST, Lou JP, Xu B, Wang J, Wang X, Liu H, Li SK, Zhen P, Zhang T
- 3261** Woman diagnosed with obsessive-compulsive disorder became delusional after childbirth: A case report
Lin SS, Gao JF
- 3268** Emphysematous pyelonephritis: Six case reports and review of literature
Ma LP, Zhou N, Fu Y, Liu Y, Wang C, Zhao B
- 3278** Atypical infantile-onset Pompe disease with good prognosis from mainland China: A case report
Zhang Y, Zhang C, Shu JB, Zhang F
- 3284** *Mycobacterium tuberculosis* bacteremia in a human immunodeficiency virus-negative patient with liver cirrhosis: A case report
Lin ZZ, Chen D, Liu S, Yu JH, Liu SR, Zhu ML
- 3291** Cervical aortic arch with aneurysm formation and an anomalous right subclavian artery and left vertebral artery: A case report
Wu YK, Mao Q, Zhou MT, Liu N, Yu X, Peng JC, Tao YY, Gong XQ, Yang L, Zhang XM
- 3297** Dedifferentiated chondrosarcoma of the middle finger arising from a solitary enchondroma: A case report
Yonezawa H, Yamamoto N, Hayashi K, Takeuchi A, Miwa S, Igarashi K, Morinaga S, Asano Y, Saito S, Tome Y, Ikeda H, Nojima T, Tsuchiya H

- 3306** Endoscopic-catheter-directed infusion of diluted (-)-noradrenaline for atypical hemobilia caused by liver abscess: A case report
Zou H, Wen Y, Pang Y, Zhang H, Zhang L, Tang LJ, Wu H
- 3313** *Pneumocystis jiroveci* pneumonia after total hip arthroplasty in a dermatomyositis patient: A case report
Hong M, Zhang ZY, Sun XW, Wang WG, Zhang QD, Guo WS

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Endoscopic-catheter-directed infusion of diluted (-)-noradrenaline for atypical hemobilia caused by liver abscess: A case report

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Abstract

BACKGROUND

Hemobilia occurs when there is a fistula between hepatic blood vessels and biliary radicles, and represents only a minority of upper gastrointestinal hemorrhages. Causes of hemobilia are varied, but liver abscess rarely causes hemobilia and only a few cases have been reported. Here, we present a case of atypical hemobilia caused by liver abscess that was successfully managed by endoscopic hepatobiliary intervention through endoscopic retrograde cholangiopancreatography (ERCP).

CASE SUMMARY

A 54-year-old man presented to our emergency department with a history of right upper quadrant abdominal colic and repeated fever for 6 d. Abdominal sonography and enhanced computed tomography revealed that there was an abscess in the right anterior lobe of the liver. During hospitalization, the patient developed upper gastrointestinal bleeding. Upper gastrointestinal endoscopy revealed a duodenal ulcer bleeding that was treated with three metal clamps. However, the hemodynamics was still unstable. Hence, upper gastrointestinal endoscopy was performed again and fresh blood was seen flowing from the ampulla of Vater. Selective angiography did not show any abnormality. An endoscopic nasobiliary drainage (ENBD) tube was inserted into the right anterior bile duct through ERCP, and subsequently cold saline containing (-)-noradrenaline was infused into the bile duct lumen through the ENBD tube with no

episode of further bleeding.

CONCLUSION

Hemobilia should be considered in the development of liver abscess, and endoscopy is essential for diagnosis and management of some cases.

Key Words: Hemobilia; Liver abscess; Noradrenaline; Endoscopic retrograde cholangiopancreatography; Case report

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Core Tip: Hemobilia occurs when there is a communication between intrahepatic blood vessels and biliary radicles caused by injury or some diseases, and represents only a minority of upper gastrointestinal hemorrhages. Liver abscess rarely causes hemobilia and only a few cases have been reported. Here, we present a rare case of atypical hemobilia caused by liver abscess that was successfully managed by catheter-directed infusion of diluted (-)-noradrenaline with an endoscopic nasobiliary drainage tube.

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INTRODUCTION

Hemobilia is defined as hemorrhage in the biliary system due to an abnormal communication between intrahepatic blood vessels and bile ducts. It represents only a minority of upper gastrointestinal hemorrhages[1]. Right upper quadrant colicky abdominal pain, obstructive jaundice, hematemesis and melena are the classic clinical presentations of hemobilia, described as Quincke's triad. There are many possible causes of hemobilia. Recently iatrogenic injury has accounted for more than half of hemobilia cases, other cases have traumatogenic, neoplastic, inflammatory, vascular causes and some are caused by gallstones[2]. Liver abscess can also cause hemobilia, although the frequency is low[3]. Here, we present a rare case of hemobilia caused by liver abscess that was successfully managed by catheter-directed infusion of diluted (-)-noradrenaline with an endoscopic nasobiliary drainage (ENBD) tube. So far, there are no reports about this treatment for intrahepatic hemobilia caused by liver abscess. Therefore, we believe that this particular case can provide an alternative option for the management of hemobilia.

CASE PRESENTATION

Chief complaints

A 54-year-old man was admitted with right upper quadrant abdominal colic and repeated fever.

History of present illness

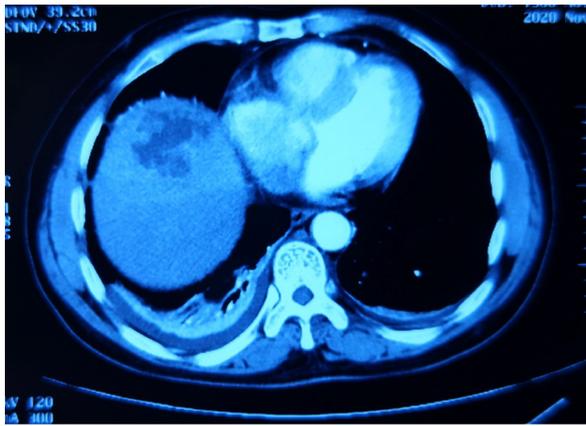
The patient developed right upper abdominal pain and repeated fever 7 d ago, and the symptoms were not relieved after treatment in the local hospital. Therefore, he was transferred to our hospital for further diagnosis and treatment.

History of past illness

The patient had a medical history of peptic ulcer and there was no past history of cholangitis or obstructive jaundice.

Imaging examinations

Enhanced computed tomography (CT) scan showed a patchy slightly low-density image (an abscess) at the top of the liver and gallstone (Figure 1). Abdominal sonography revealed that there was an abscess in the right anterior lobe of the liver.



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Figure 1 Enhanced computed tomography scan of the abdomen revealed patchy slightly low-density image at the top of the liver.

Laboratory examinations

Initial laboratory investigation presented hemoglobin 107 g/L, white blood cell count $13.11 \times 10^9/L$, platelet count $206 \times 10^9/L$, serum albumin 34.2 g/L, alanine aminotransferase 43 IU/L, glutamic oxalacetic transaminase 31 IU/L, alkaline phosphatase 232.6 IU/L, γ -glutamyl transpeptidase 231.3 IU/L, total bilirubin 18.3 $\mu\text{mol/L}$, direct bilirubin 9.7 $\mu\text{mol/L}$, and hypersensitive C-reactive protein 73.77 mg/L. Coagulation profiles were within normal limits, but fecal occult blood was weakly positive. In addition, detection of ameba, enteric typhoid and rotavirus was also negative.

Physical examination

On admission, he had normal temperature with blood pressure of 124/86 mmHg and pulse rate of 86 beats/min. Physical examination revealed that there was moderate tenderness in the right upper quadrant of the abdomen with no rebound tenderness or Murphy's sign, and no jaundice of skin and sclera.

Personal and family history

The patient had no family history that was related to the present illness.

FINAL DIAGNOSIS

Diagnosis of liver abscess and the possibility of upper gastrointestinal bleeding.

TREATMENT

The patient received ultrasound-guided percutaneous (catheter) drainage of the abscess cavity and was given cephalosporin antibiotics and hemostatic drugs when he completed the examination. On day 2 of hospitalization, the patient had three episodes of melena and one of syncope for a few seconds. His hemoglobin decreased to 60 g/L with unstable hemodynamics demonstrated by increased pulse rate of 103 beats/min and declined blood pressure of 90/63 mmHg. Immediate resuscitation was carried out with infusion of 6 U packed red cells and 400 mL fresh frozen plasma. Upper gastrointestinal endoscopy revealed duodenal ulcer bleeding, and three metal clamps were applied to stop the bleeding. After a brief period of hemodynamic stabilization, hemoglobin levels dropped to 63 g/L. Therefore, upper gastrointestinal endoscopy was performed once again and fresh blood was seen flowing from the ampulla of Vater (Figure 2). However, selective angiography did not show any abnormality (Figure 3A). Endoscopic retrograde cholangiopancreatography (ERCP) indicated no filling defects in the intrahepatic and extrahepatic bile ducts, but when the guide-wire entered the marginal bile duct of the right anterior branch, a large amount of bloody bile was seen spilling from the ampulla of Vater. An ENBD tube was inserted into the right anterior bile duct, and subsequently cold saline containing (-)-noradrenaline (0.9% saline 100 mL with 8 mg (-)-noradrenaline) was infused into the bile duct lumen through the ENBD tube (10 mL/h) (Figure 3B).



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Figure 2 Endoscopic nasobiliary drainage revealed fresh blood flowed from the ampulla of Vater.



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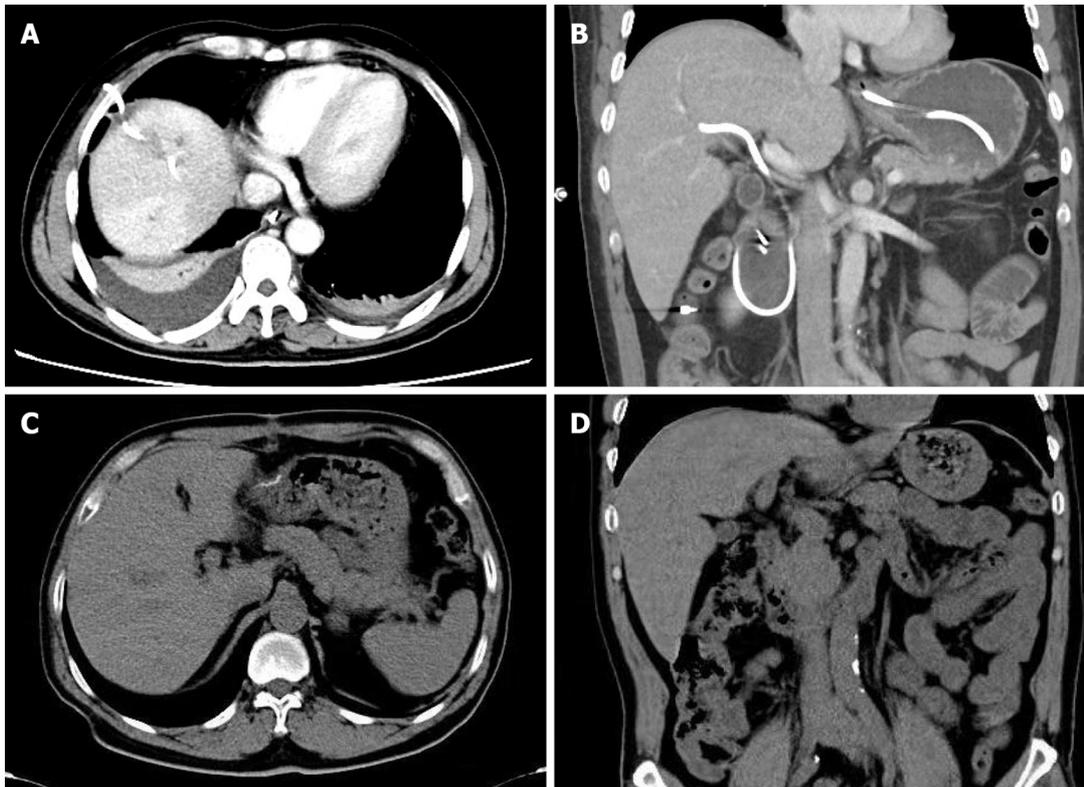
Figure 3 Angiography. A: Selective hepatic artery angiography did not show active bleeding; B: Endoscopic nasobiliary drainage tube was placed in the right anterior bile duct to implement catheter-directed infusion of diluted (-)-noradrenaline.

OUTCOME AND FOLLOW-UP

There was no episode of further bleeding, and abdominal CT scan performed on postoperative day 10 showed that the abscess had basically disappeared (Figure 4A and B). The patient was discharged without any complications. One month later, abdominal CT revealed no expansion of the intrahepatic bile duct and complete absorption of the liver abscess (Figure 4C and D).

DISCUSSION

Hemobilia represents only a minority of upper gastrointestinal hemorrhages and occurs when there is a fistula between intrahepatic blood vessels and biliary radicles. The etiology of hemobilia is diverse. It can have traumatogenic causes; iatrogenic causes (percutaneous liver procedures, hepatobiliary surgery and endoscopic hepatobiliary procedures); or noniatrogenic causes (cholelithiasis, inflammatory diseases, calculous cholecystitis, cholangitis, and parasitic infection); and it may be due to vascular disorders (aneurysms); neoplasms and coagulopathy[4]. In recent years, because of the increasing popularity of interventional diagnosis and treatment of the hepatobiliary tract, most cases of hemobilia



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Figure 4 Abdominal computed tomography scan. A and B: Abdominal computed tomography scan was performed on postoperative day 10. Liver abscess was absorbed by the ultrasound-guided percutaneous catheter drainage (A); Endoscopic nasobiliary drainage tube was placed in the right anterior bile duct without displacement (B); C and D: Abdominal computed tomography scan was performed 1 mo later. There was no intrahepatic mass found (C); There was no expansion of the bile duct inside and outside the liver (D).

have been caused by iatrogenic injury[4]. Although liver abscess can also cause hemobilia, only a few cases have been reported[3-5]. At present, the specific pathological mechanism of hemobilia caused by liver abscess is still unclear, and the possible explanation is erosion or segmental ischemia of the distal bile duct wall by abscess[5].

The classic clinical presentation of hemobilia is described as Quincke's triad of right upper quadrant colicky abdominal pain, obstructive jaundice, hematemesis and melena. However, only 22%-35% of cases present with the triad, and even some cases manifest clinical silence[1]. On admission, our patient presented signs of infection supported by the laboratory findings that were compatible with liver abscess. Only a weakly positive test for fecal occult blood suggested the possibility of gastrointestinal bleeding. The patient developed hemodynamic instability and melena on the second day of hospitalization. During the clinical course of hemobilia, he did not manifest obstructive jaundice because there were no intraductal blood clots to induce biliary stasis, and the formation of clots may be related to the location, amount and rate of bleeding.

Diagnosis of hemobilia is challenging because it is uncommon, especially in some cases where there are no risk factors such as recent biliary tract manipulation or trauma. Imaging techniques such as ultrasound and computed tomography can be helpful for the initial diagnosis and guiding the choice of treatment, although the findings are often indirect and nonspecific[6]. Upper endoscopy is the most frequent way to provide direct visible evidence of bleeding from the ampulla of Vater, particularly in suspected cases of hemobilia, and repeated upper endoscopy or side-viewing duodenoscopy should be considered[7]. Until recently, conventional angiography was not recommended as a first-line management because of its invasive nature, and angiographic embolization can only be used if the bleeding is vigorous or if vascular abnormalities (aneurysm or pseudoaneurysm) are visible[8,9]. Surgical intervention plays a complementary role in the treatment of hemobilia in cases of failed embolization[9]. In the present case, repeated abdominal ultrasound examination at admission did not reveal bile duct expansion inside or outside the liver, and there was only a strong echo vocal shadow in the gallbladder cavity. We initially misinterpreted upper gastrointestinal bleeding from ulcer bleeding due to peptic ulcer. A second endoscopy finally revealed fresh blood leak from the ampulla of Vater. However, selective angiography did not show any abnormality, and it was suspected that the bleeding originated from a portal vein or hepatic vein branch. ERCP was performed to manage hemostasis through a variety of endoscopic techniques and accessories, which depends on the cause, location and vascular source[10,11]. Because the abscess was located in the right lobe of the liver, and when a guide-

wire entered the marginal bile duct of the right anterior branch, a large amount of bloody bile was seen spilling from the ampulla of Vater. In light of this situation, we speculated that the bleeding site was located in the right anterior bile duct, thus an ENBD tube was placed to implement catheter-directed infusion of diluted (-)-noradrenaline. This technique offers unique benefits and it is effective and safe especially for submucosal venous hemorrhage of the bile duct[12], but it is not carried out routinely mainly due to related discomfort, technically cumbersome procedures and easy inadvertent catheter withdrawal[13]. Hemobilia caused by liver abscess in our patient was obviously cured without further operative intervention.

CONCLUSION

The possibility of hemobilia should be considered in the development of liver abscess although the frequency of the complication is low. Endoscopic-catheter-directed infusion of hemostatics *via* ENBD tube is an effective and safe conservative treatment for hemobilia caused by liver abscess.

FOOTNOTES

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