Non-Operative Management of Bronchobiliary Fistula Due to Proximal Migration of Biliary Stent in a Patient With Unresectable Klatskin Tumor

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ABSTRACT

INTRODUCTION: One of the methods for the biliary tree decompression in the case of Klatskin tumor is transpapillary stenting, which could be completed by stent migration in 4% to 10% of cases. Approximately half of the stent migrations are in the proximal direction. In this study, we reported a rare case of proximal trans-diaphragmatic stent migration to the lower lobe of the right lung with the formation of a biliary-bronchial fistula (BBF).

CASE PRESENTATION: A 60-year-old woman was diagnosed with hilar cholangiocarcinoma (type 3B by Bismuth-Corlette) complicated by posthepatic jaundice. To relieve jaundice there were performed endoscopic retrograde cholangiopancreatography, endoscopic sphincterotomy, endobiliary stent placement (10 Fr, 150 mm). A restenting (11.5 Fr, 130 mm) was performed in 2.5 months due to endobiliary tube occlusion. In the next 2 months, coughing attacks and biliptysis have appeared in the patient. A CT scan showed penetration of the liver, diaphragm, and lower lobe of the right lung with the proximal part of the stent and caused BBF formation. Anti-inflammatory and antibacterial therapy was administrated for 14 days and BBF was closed. Stent retrieval from the right hemithorax and endobiliary restenting was performed in 9 months after primary stenting. During follow-up, appropriate positioning and functioning of the stent were observed.

CONCLUSION: BBF formation is a rare complication of endobiliary stenting, which can be successfully treated by anti-inflammatory and antibiotic therapy, followed by transpapillary stent retrieval.

KEYWORDS: Hilar cholangiocarcinoma, Klatskin tumor, endoscopic retrograde cholangiopancreatography, ERCP, biliary-bronchial fistula, endobiliary stent, proximal migration of stent, non-operative treatment

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Introduction

Biliary-bronchial fistula (BBF) is a rare disorder, which is reported mainly in patients after hepatic resections.[1] BBF was also reported as a complication after surgical treatment for hilar cholangiocarcinoma, also known as Klatskin tumor. Klatskin tumor is a rare malignant neoplasm, arising from the hepatic ducts and causing posthepatic jaundice. To treat this malignancy, surgical resection is applied to excise the primary tumor, whereas placement of the stent is performed to achieve biliary decompression as a palliative measure.[2] The rate of biliary stent migration is 4% to 10% and stent migration into proximal directions is rare event.[2–6] Still, the migrated stent may perforate the diaphragm, causing the subsequent formation of the BBF, presenting with such symptoms as biliptysis and severe bronchopneumonia. The aim of this study was to present a rare case of successful non-operative management of BBF in a patient with an unresectable Klatskin tumor.

Case Report

A 60-years-old female was admitted with signs of posthepatic jaundice and coagulopathy: total bilirubin (TB) — 538.7 µmol/L, direct bilirubin (DB) — 412.1 µmol/L. Coagulation test showed fibrinogen level at 110 mg/dl (normal range 180-350 mg/dl), prothrombin Quick value — 46% (normal range 0.85-1.2), international normalized ratio (INR) — 2.4 (normal range 0.85-1.2), indicating high risk of bleeding. A diagnosis of Klatskin tumor of the left hepatic duct (type 3B by Bismuth-Corlette classification) was established based on a contrast-enhanced CT scan and biopsy. The CT scan analyses revealed the tumor to be intimately adherent to the common hepatic artery (highly suspicious for invasion) as well as wrapped around the half of the portal vein. The resection of the liver with major vessels was not performed because such operation is not covered by the Medical Guarantees Package from the National Health Service of Ukraine due to poor financing of healthcare in Ukraine,[7] and the patient was not able to cover such expenses by herself. The malignancy was diagnosed as an

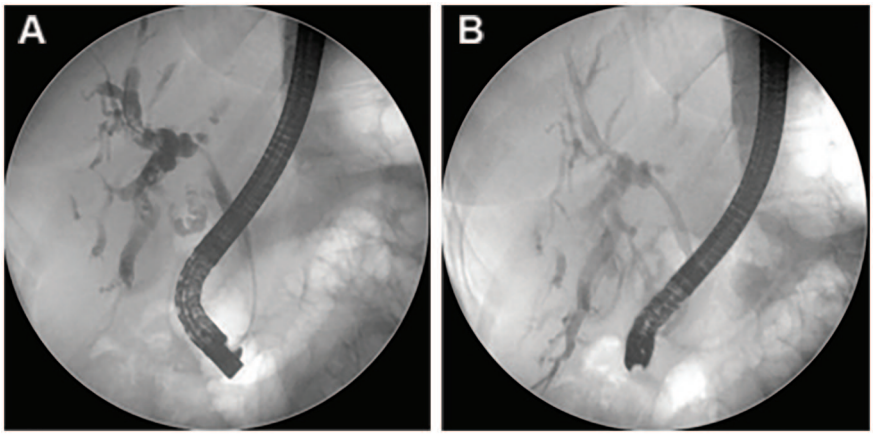

Figure 1. ERCP in tight filling phase. The wire conductor was inserted into the posterior sectional bile duct through the right hepatic duct: (A) the dilatation of the intrahepatic bile ducts up to 11 mm was determined and (B) the endobiliary PVC stent 10 Fr 150 mm was installed into the right posterior sectional bile duct followed by free passage of contrast medium into the duodenum.

unresectable tumor; therefore, a palliative minimally invasive approach was chosen. The study was approved by the local ethical committee at Bogomolets National Medical University. The patient provided written informed consent to participate in the study. All data generated or analyzed during this study are included in this published article.

Initially, we thought that percutaneous transhepatic biliary drainage might be favorable compared to the endoscopic approach. However, the decision was made to performed an endoscopic approach because the patient had a Klatskin tumor with a spread into the left hepatic duct, whereas the right hepatic duct was dilated and free from tumor, indicating a good prognosis for endoscopic management in this particular case. The dilation of the right hepatic duct was due to obstruction at the level of bile duct confluence. Also, a high risk of bleeding was taken into consideration, and a decision was made to perform endoscopic retrograde cholangiopancreatography (ERCP) followed by endoscopic sphincterotomy and endobiliary stenting. Application of self-expanding metal stents (SEMS) is associated with better clinical outcomes and enhanced biliary decompression. As compared to plastic stents, SEMS have a wider diameter, which is resulted in enhanced bile flow after the stent insertion. In such a condition, biliary decompression is rapid, and the risk of liver failure is increased according to Khoronko et al[8] and our clinical observations (unpublished data). Hence, we decided to use a plastic stent in order to achieve gradual decompression because of signs of liver failure due to posthepatic jaundice. The polyvinyl chloride (PVC) stent (10 Fr, 150 mm) was placed into the right hepatic duct (Figure 1) and colorless "white" bile was received. The selective stenting of the right hepatic duct was achieved by maneuvering the flexible part of the endoscope and its elevator as well as using guidewires with J-tip, fixed, and moveable core. Also, a significant decrease in bilirubin levels was achieved. In 2.5 months after the PVC insertion, the patient was diagnosed with posthepatic jaundice and cholangitis: TB — 120.6 μmol/L, DB — 86.3 μmol/L, WBC — 12.7×10^9/L, band neutrophils (BN) — 18%, erythrocyte sedimentation rate (ESR) — 29 mm/hour. Occlusion of the PVC stent was established by MRI, followed by ERCP (Figure 2), and replacement of the stent was performed with a larger diameter (11.5 Fr, 130 mm). The patient was administrated chemotherapy with 3 g of capecitabin (18 courses).

Two months later after the endobiliary restenting procedure, the patient reported fever up to 38°C and dry cough with a sudden onset. According to the patient, she had an active lifestyle, doing regular chores like farming and gardening in the countryside. During the next 7 days after the initial presentation, the dry cough was changed to a wet one with an admixture of bile, indicating biliptysis. The blood test showed WBC — 10.5×10^9/L, ESR — 66 mm/hour, TB — 12 μmol/L. CT scan showed the proximal migration of the endobiliary stent with penetration through the liver, the right dome of the diaphragm, and right lung (Figure 3). The distal part of the stent was at the level of the common hepatic duct (Figure 3C and D). The patient was not able to receive immediate endoscopic management, because such a clinical facility was not available in the area of her residence, and transportation to a tertiary referral center was not possible because of patient's fear to get infected with COVID-19. In such circumstances, conservative therapy was started by administration of Meloxicam 15 mg/day, Amoxicillin 1.5 g/day, Ceftazidim 3 g/day, and Furosemide 40 mg/day. The normalization of body temperature and blood parameters as well as the absence of biliptysis were reported in 2 weeks.

During the follow-up of 9 months, there were detected neither signs of BBF nor posthepatic jaundice. MRI investigation showed signs of fibrotic tissue around the part of the endobiliary stent in the pulmonary parenchyma with a clear demarcation from the normal lung tissue (Figure 4).

As the epidemic situation with COVID-19 became better, the patient arrived at a tertiary center for endoscopic treatment in Kyiv (National Military Medical Clinical Center of Ministry of Defense of Ukraine). To remove the proximally

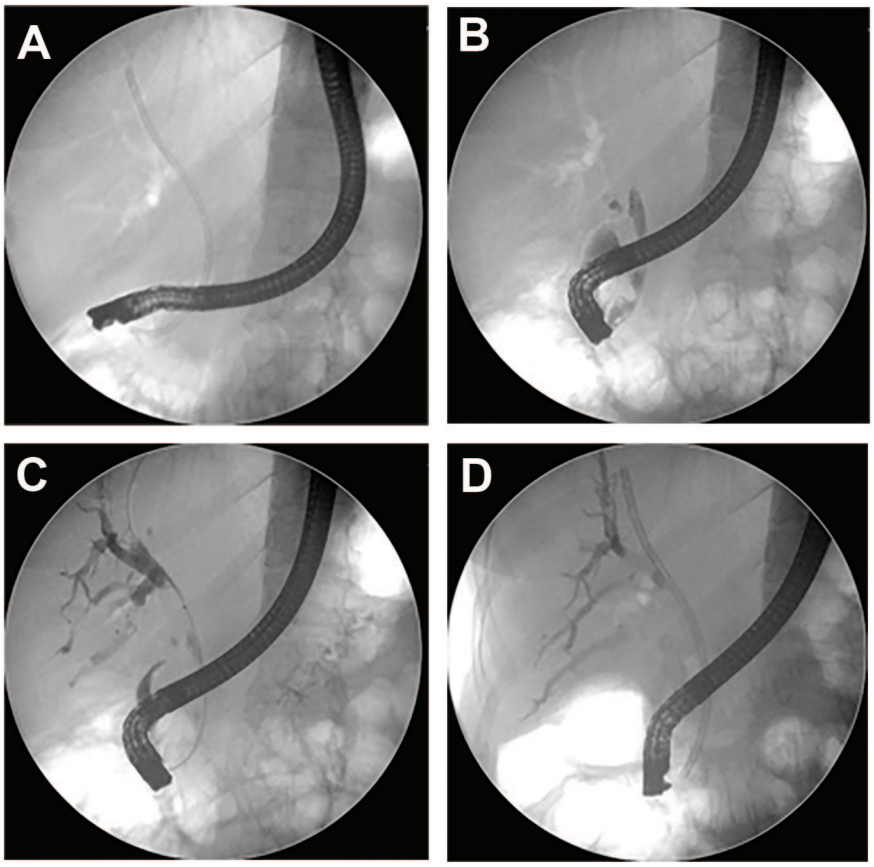

Figure 2. Illustration of restenting stages by ERCP of the previously installed and completely occluded endobiliary 10 Fr stent: (A) ERCP in the tight filling phase showed absence of the contrast passage to the common bile duct indicating occlusion of the stent. (B) The wire conductor was introduced to the right hepatic and the posterior sectional bile ducts. The dilated common hepatic duct and proximal bile ducts were filled with contrast medium. (C) The PVC endobiliary stent 11.5 Fr 130 mm was placed to the right posterior sectional bile duct. (D) Illustration of free passage of contrast medium into the duodenum.

migrated endobiliary stent we visualized its proximal tip by ERCP in the common bile duct at the level of the cystic duct. Then, the tip was captured with 4 wire Dormia basket followed by its removal. The removed stent was replaced with a PVC stent (10 Fr, 150 mm) by ERCP (Figure 5). The surgical team considered possible technical problems and complications associated with fibrosis around the migrated stent, and conversion to open surgery was considered in case of unsuccessful endoscopic management. Complications were not observed at follow-up.

Discussion

In this study, we reported a case of nonoperative management of a rare BBF in the patient with a Klatskin tumor. A biliary fistula is usually associated with injury to the liver in civil and combat trauma to be complicated by external bile leak, whereas BBF is considered as a rare biliary complication.[9–12]

In this report, we have shown a BBF as a complication after the palliative stenting in the patient with malignant neoplasm, which is in line with other studies and reviews.[2,3,5,10,12] Our findings are also in line with Gandini et al,[10] demonstrating a case of BBF in the patient with Klatskin tumor after the hepatic surgery and with signs of irritating cough with biliptysis and fever. Similar clinical findings and more severe distress syndrome and pneumonia were also reported by Antonsen et al,[13] showing a case of BBF due to proximal stent migration from the right hepatic duct after traumatic necrosis of the common bile duct. In contrast to the studies of non-operative BBF management, Joh and Park[14] reported pulmonary resection and omentopexy to treat BBF. In our observation, a decision was made against the surgical operation, since conservative treatment was effective. Still, the question remains unclear about the urgent retrieval of the endobiliary stent upon the diagnosis of BBF. For example, Antonsen et al[13] reported a lethal outcome after retrieval of the stent before the anti-inflammatory drug treatment, whereas other studies advocated non-operative management, which is demonstrated a good result and could be considered as an acceptable therapeutic approach.[1,5,10] In our case, we applied a non-operative approach for the retrieval of the migrated stent, which is in line with other case reports as well as large studies from tertiary level endoscopy centers.[15,16] The operative techniques are used in minor cases due to high risk of surgery complications or as unexpected findings.[16,17]

It is worth to mention, that other possible causes of BBF were described such as trans-diaphragmatic migration of a

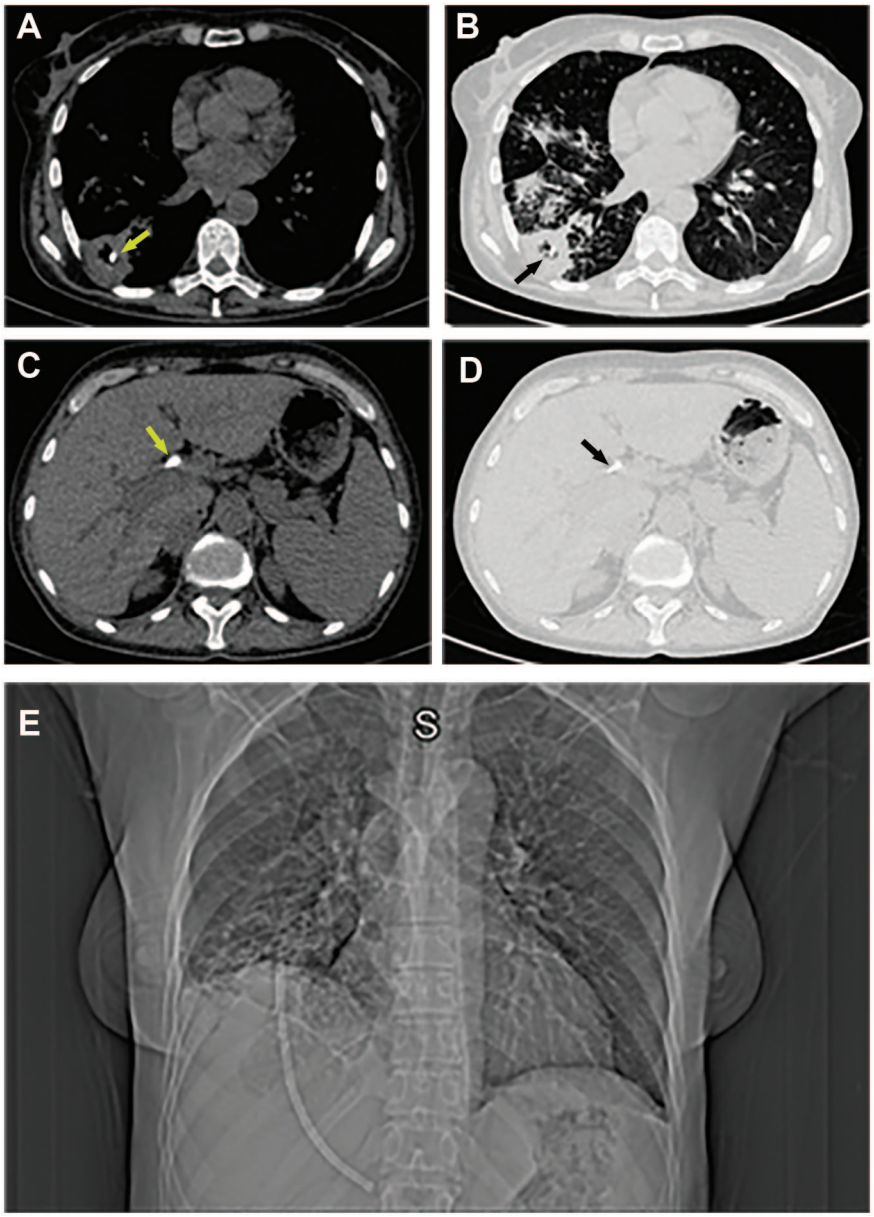

Figure 3. Chest and abdomen CT scan: A and C—Routine 1.5 B31s; B and D—Routine 1.5 B70s; E—Topogram 0.6 T20f. (A and B) The proximal end of the stent is located in the posterior part of the lower lobe of the right lung (indicated by an arrow), surrounded by an infiltration area. (C and D) The distal end of the endobiliary stent (indicated by an arrow) is displaced into the common hepatic duct.

biliary stent due to iatrogenic injury as well as biliary obstruction and lung abscess in a patient with pancreatic neuroendocrine tumor.[9,18]

In the majority of cases, the clear mechanism of proximal trans-diaphragmatic migration of the endobiliary stent with the formation of BBF remains controversial. Richenberg et al[15] demonstrated plastic stent migration 14 days after its application and referred to the study of Johanson et al[19] suggesting such possible causes of the stent migration as the presence of malignant strictures, short stents with large diameter. Our experience led to the hypothesis that trans-diaphragmatic stent migration is associated with a subphrenic abscess formation due to angiotrophic necrosis of the adjacent liver and diaphragm tissue in cases of long-term presence of the endobiliary stent. It is also worth to mention, that a possible negative role for the BBF formation may play chemotherapy, which is associated with the impaired reparative capacity of the tissues. We also hypothesize that predisposing factors for the stent migration were dilated and thinned bile ducts, a large stent diameter (11.5 Fr), palliative chemotherapy for Klatskin tumor, possible malignant strictures as well as patient's high physical activity after stenting. However, these factors cannot be exclusively contributed to such an early (within 2 months) trans-diaphragmatic migration of the stent to the right lung. Here we propose that a possible mechanism for the early stent migration may associate with dysfunction of the special retainers at the edges of a stent. Thus, during minimal accidental proximal stent dislocations, the proximal retainers came into firmer contact with the sectional bile duct as compared to the distal retainers, which are located in the common

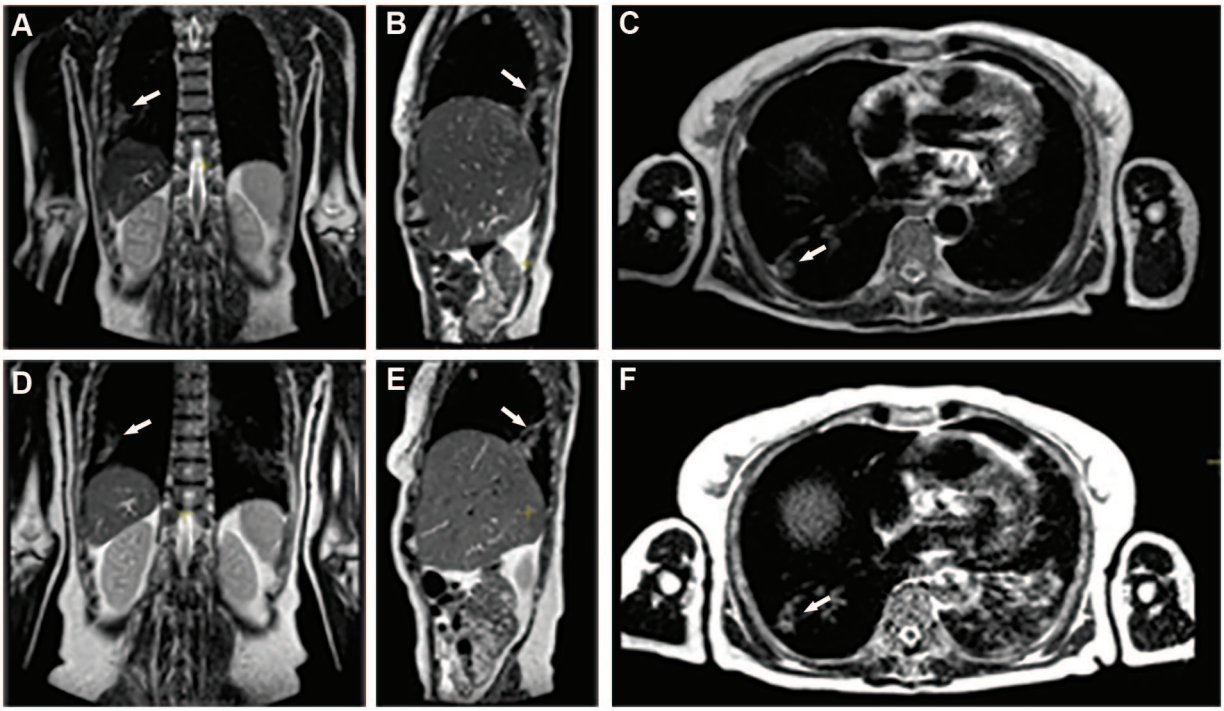

Figure 4. Chest and abdomen MRI follow-up showing the presence of biliary-bronchial fistula (BBF). Arrows indicate the bronchial portion of BBF with signs of perifocal pneumonsclerosis in both images. (A–C) MRI signs of BBF at 4 months after the diagnosis. (D–F) MRI signs of BBF at 9 months after the diagnosis.

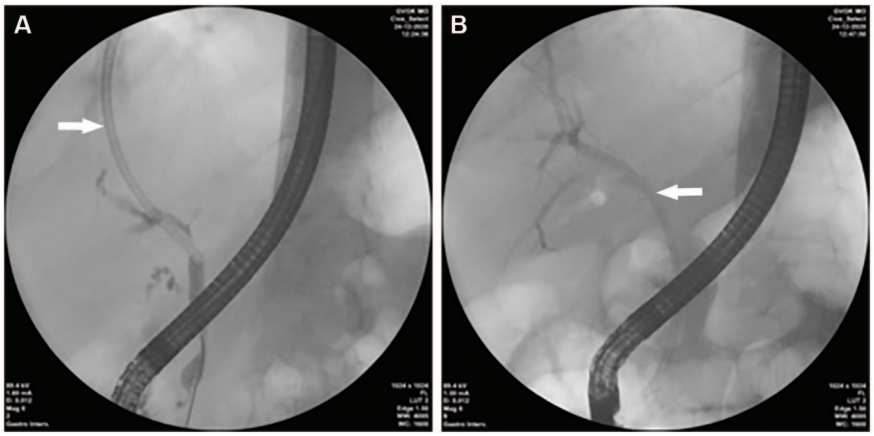

Figure 5. Illustration of ERCP findings: (A) The X-ray image before retrieval of the migrated endobiliary stent 11.5 Fr 130 mm (indicated by an arrow). The proximal part of the stent was migrated to the common hepatic duct, whereas the distal end was migrated to the lower lobe of the right lung. (B) The X-ray image after removal of the migrated stent and re-draining right hepatic duct with a new stent 10 Fr 150 mm (indicated by an arrow).

biliary duct. As a result of such a contact, a stent was engaged to move only in an incremental unidirectional (proximal) way resulting in the perforation of adjacent tissues. This hypothesis is in line with Bascom et al.[20] and Lurin and Tsema[21] showing similar pathogenetic features in studies of pilonidal sinus formation due to the trichogenic-pump mechanism.

Conclusion

BBF formation is a rare complication of endobiliary stenting, which should be considered in routine clinical settings in patients with hepatic tumors. In some cases, non-operative management of BBF is associated with a good outcome, showing excellent compliance with anti-inflammatory and antibacterial therapy with transpapillary extraction of the proximally migrated stent.

Authors' contributions

IT – acquisition of data, analysis and interpretation of data, literature search, drafting of manuscript; VS – acquisition of data, analysis and interpretation of data; DR – acquisition of data, analysis and interpretation of data; DM – analysis and interpretation of data, critical revision, drafting of manuscript,

literature search and analyses, drafting of manuscript; OY - acquisition of data, analysis and interpretation of data, preparing of figures, literature search; AD – study conception, critical revision of manuscript.

Consent for Publication

For this type of study consent for publication is not required.

Data Access Statement

The authors confirm that the data supporting the findings of this study are available within the article.

Ethical Approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. The study was approved by the local ethical committee at Bogomolets National Medical University.

Informed Consent

Informed consent was obtained from individual participant included in the study.

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REFERENCES

1. Ammirabile A, Rizzo DA, Pedicini V, Lanza E. Percutaneous treatment of bronchobiliary fistula: report of a successful transhepatic embolization and a decision-making strategy driven by systematic literature review. *Cardiovasc Intervent Radiol*. 2021;44:1005-1016.
2. Taj MA, Ghazanfar S, Qureshi S, Zubair M, Niaz SK, Quraishy MS. Plastic stent migration in ERCP; a tertiary care experience. *J Pak Med Assoc*. 2019;69:1099-1102.
3. Katsinelos P, Kountouras J, Paroutoglou G, et al. Migration of plastic biliary stents and endoscopic retrieval: an experience of three referral centers. *Surg Laparosc Endosc Percutan Tech*. 2009;19:217-221.
4. Arhan M, Odemiş B, Parlak E, Ertuğrul I, Başar O. Migration of biliary plastic stents: experience of a tertiary center. *Surg Endosc*. 2009;23:769-775.
5. Tarnasky PR, Cotton PB, Baillie J, et al. Proximal migration of biliary stents: attempted endoscopic retrieval in forty-one patients. *Gastrointest Endosc*. 1995;42:513-520.
6. Mohammad Alizadeh AH, Donboli K, Khodakarami M, Baghbani S, Zali MR. Biliary stent migration to hepatic duct-case report of a late complication. *Clin Med Insights Case Rep*. 2015;8:13-14.
7. Dinets A, Nykytiuk O, Gorobeiko M, Barabnchyk O, Khrol N. Milestones and pitfalls in strategic planning of healthcare in capital city in transition. *Georgian Med News*. 2021;315:189-195.
8. Khoronko YV, Korobka VL, Groshilin VS, Tolstopyatov SV, Shitikov IV. "Rapid" biliary decompression syndrome in obstructive jaundice surgery (In Russian). *Annaly khirurgicheskoy gepatologii = Ann HPB Surg*. 2019;24:123-129.
9. Dasmahapatra HK, Pepper JR. Bronchopleurobiliary fistula. A complication of intrahepatic biliary stent migration. *Chest*. 1988;94:874-875.
10. Gandini R, Konda D, Tisone G, Pipitone V, Anselmo A, Simonetti G. Bronchobiliary fistula treated by self-expanding ePTFE-covered nitinol stent-graft. *Cardiovasc Intervent Radiol*. 2005;28:828-831.
11. Khomenko I, Tsema I, Humeniuk K, et al. Application of damage control tactics and transpapillary biliary decompression for organ-preserving surgical management of liver injury in combat patient. *Mil Med*. Published online April 16, 2021. doi:10.1093/milmed/usab139
12. Liao GQ, Wang H, Zhu GY, Zhu KB, Lv FX, Tai S. Management of acquired bronchobiliary fistula: a systematic literature review of 68 cases published in 30 years. *World J Gastroenterol*. 2011;17:3842-3849.
13. Antonsen J, Preisler L, Skram U, Klein M. Fatal biliary-pulmonary fistula due to a proximally migrating biliary stent. *Endoscopy*. 2013;45 Suppl 2 UCTN: E265-E266.
14. Joh HK, Park SY. Surgical treatment of bronchobiliary fistula with pulmonary resection and omentopexy. *Korean J Thorac Cardiovasc Surg*. 2020;53:38-40.
15. Richenberg JI, Amin Z, Hatfield AR, Lees WR. Pleurobiliary fistula following Tannenbaum biliary stent insertion. *Eur Radiol*. 1996;6:697-699.
16. Lahoti S, Catalano MF, Geenen JE, Schmalz MJ. Endoscopic retrieval of proximally migrated biliary and pancreatic stents: experience of a large referral center. *Gastrointest Endosc*. 1998;47:486-491.
17. Ozair A, Ahmad F, Kumar S, Rungta S. Transhepatic intraperitoneally migrated biliary stent: a rare finding during laparoscopic cholecystectomy. *J Minim Access Surg*. 2021;17:85-87.
18. Panda D, Aggarwal M, Yadav V, Kumar S, Mukund A, Baghmar S. Bronchobiliary fistula secondary to biliary obstruction and lung abscess in a patient with pancreatic neuro-endocrine tumor. *J Egypt Natl Canc Inst*. 2016;28:117-121.
19. Johanson JF, Schmalz MJ, Geenen JE. Incidence and risk factors for biliary and pancreatic stent migration. *Gastrointest Endosc*. 1992;38:341-346.
20. Bascom J. Pilonidal disease: origin from follicles of hairs and results of follicle removal as treatment. *Surgery*. 1980;87:567-572.
21. Lurin I, Tsema E. Etiology and pathogenesis of pilonidal disease. *Coloproctology*. 2013;3:35-49.