Elbahr *Egyptian Liver Journal* (2019) 9:4
https://doi.org/10.1186/s43066-019-0005-8

Egyptian Liver Journal

CASE REPORT Open Access

Biliptysis

Osama Elbahr

Abstract

Background: Biliptysis means coughing of bile which is a presenting symptom of a rare condition called bronchobiliary fistula (BBF). BBF is a connection between the biliary tract and bronchial tree. BBF mostly occurs secondary to malignancy, liver abscess, and trauma. Surgical approach in BBF management was the main management strategy, then endoscopic approach.

Case presentation: We managed our first encountered case of biliptysis endoscopically by endoscopic retrograde cholangiopancreatography (ERCP).

Conclusion: ERCP management seems to be effective in management of biliptysis.

Keywords: Biliptysis, Bronchobiliary, Fistula, Bile, ERCP

Background

Bronchobiliary fistulas are rare. In most cases, they are caused by neoplasms and hepatic or subphrenic abscesses, resulting from different conditions or trauma [1]. It is usually diagnosed by clinical history (coughing of bile) and imaging (CT/MRI) [2, 3]. Treatment is usually surgery or endoscopic or transhepatic embolization [4, 5]. We used an ERCP endoscope and placing of stent for appropriate closure of the fistula.

Case presentation

A 26-year-old male working as a cook in a restaurant presented with a history of fever and abdominal pain for 2 weeks and received therapy in the form of empiric antibiotics and antipyretics with no improvement. Later, the patient started to complain from coughing of dark yellowish sputum (biliptysis). A CT scan on the abdomen and lower chest was ordered and revealed large subphrenic abscess (Fig. 1). Then, the patient was referred to our hospital (National Liver Institute (NLI), Shebin Alkawm, Egypt) and was admitted to the surgery department. CT of the abdomen was repeated and confirmed the subphrenic abscess connected to the lower lung zone abscess with right hepatic lobe abscess. Aspiration of fluid sample from the abscess for bacteriological evaluation revealed pure pus with negative culture for any organism (may be related to how much antibiotics the patient had received). Liver profile was normal which is a rare finding in such hepatic lesions. Kidney function was also normal. He had negative virology markers (HBV, HCV, and HIV).

The patient was referred to our department (hepatology and gastroenterology) to give him a trial of endoscopic management. ERCP was done, and the fluoroscopic image revealed pooling of contrast in the right hepatic lobe (abscess) with contrast seen tracking upward to the lower lung (Fig. 2 is a picture of bronchobiliary fistula). Cannulating an intrahepatic small biliary duct by guidewire to bypass the site of fistula and drain the hepatic abscess was successful,

Correspondence: osama.elbahr@liver.menofia.edu.eg
National Liver Institute, Shibin Al Kawm, Menoufia, Egypt

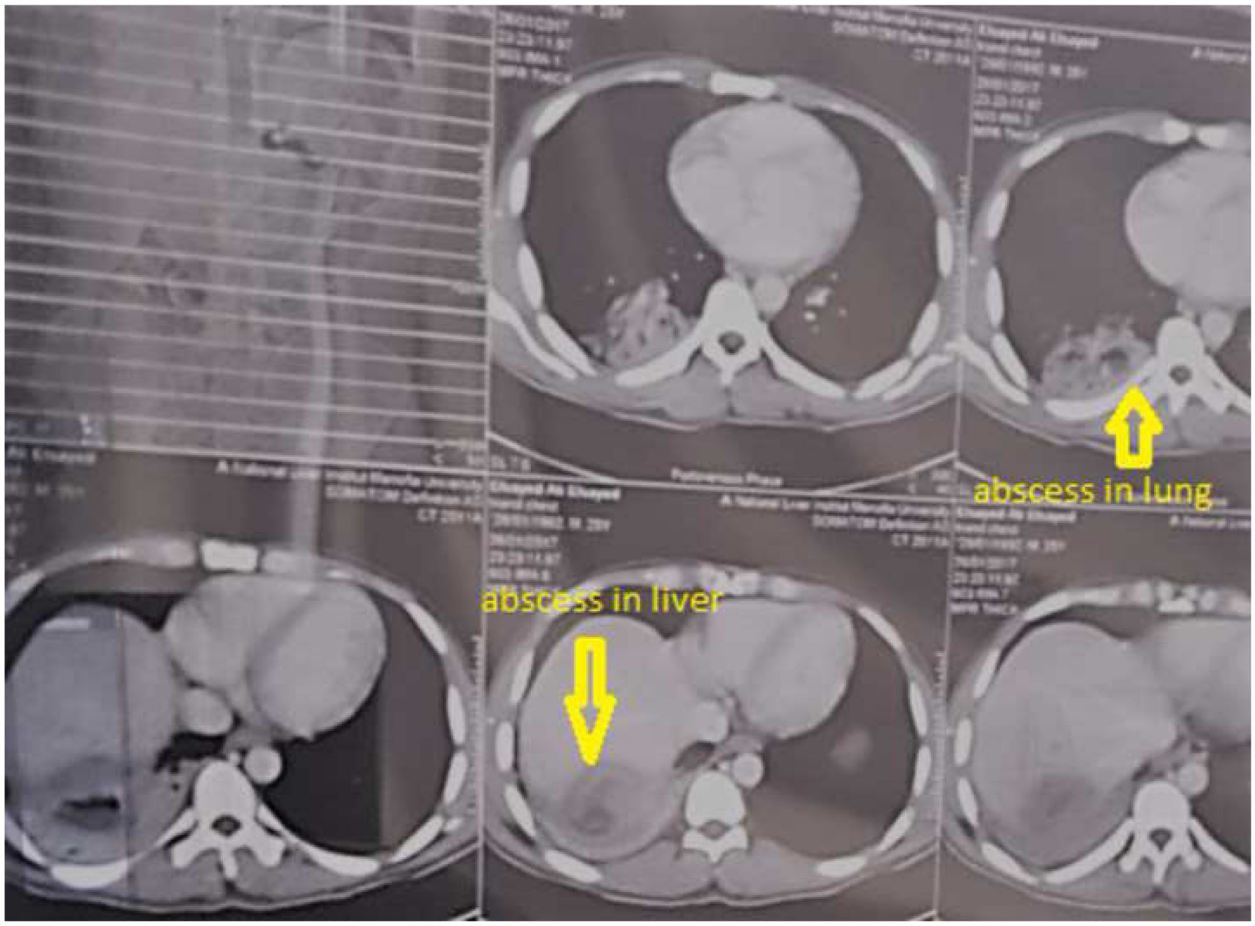

Fig. 1 CT of the lower chest and abdomen showing the abscess in the liver and lower lung

and this was followed by inserting a 10–15-cm plastic stent. Improvement of biliptysis was achieved which is associated with improvement of fever. The patient was discharged from our institute 5 days after endoscopy and was followed up at the surgery department weekly for 3 months. His symptoms totally improved, and follow-up CT showed resolution of the abscess.

BBF, despite being rare, is known to be caused by different causes: congenital, malignancies, abscesses, traumatic, or iatrogenic [4, 6]. Diagnosis is usually made clinically for biliptysis and it is sometimes inappropriately diagnosed as pneumonitis or chronic cough with greenish sputum [2] [7].

Somatostatin and its analogues were tried for treating BBF, by reducing digestive tract secretions [8]. Patients should be advised to take orthostatic position and avoid supine position to decrease the volume of coughed bile and accelerate fistula healing. Also, supporting therapy should be administered with appropriate prophylactic attention to electrolyte disturbance [2].

Definitive treatment for BBF has not yet been established. Surgical or non-surgical interventional procedures, such as endoscopic retrograde cholangiopancreatography (ERCP) or percutaneous transhepatic drainage (PTD), are frequently used as direct photographic evidence and management [9].. Transhepatic embolization, bronchoscopic injection of *n*-butyl cyanoacrylate, or histoacryl embolization has been tried [4, 7]. A systematic review done on 68 cases had reported that interventional procedures were slightly more effective than surgical procedure (97% vs. 85%) [2].

In the case of BBF due to abscess, we think two combined approaches could be attempted. The first approach is abscess management by antibiotics and drainage, and the second one is biliary drainage either by ERCP or PTD. Shrinking the abscess cavity closes the fistula tract, and biliary drainage prevents the recurrence of the fistula.

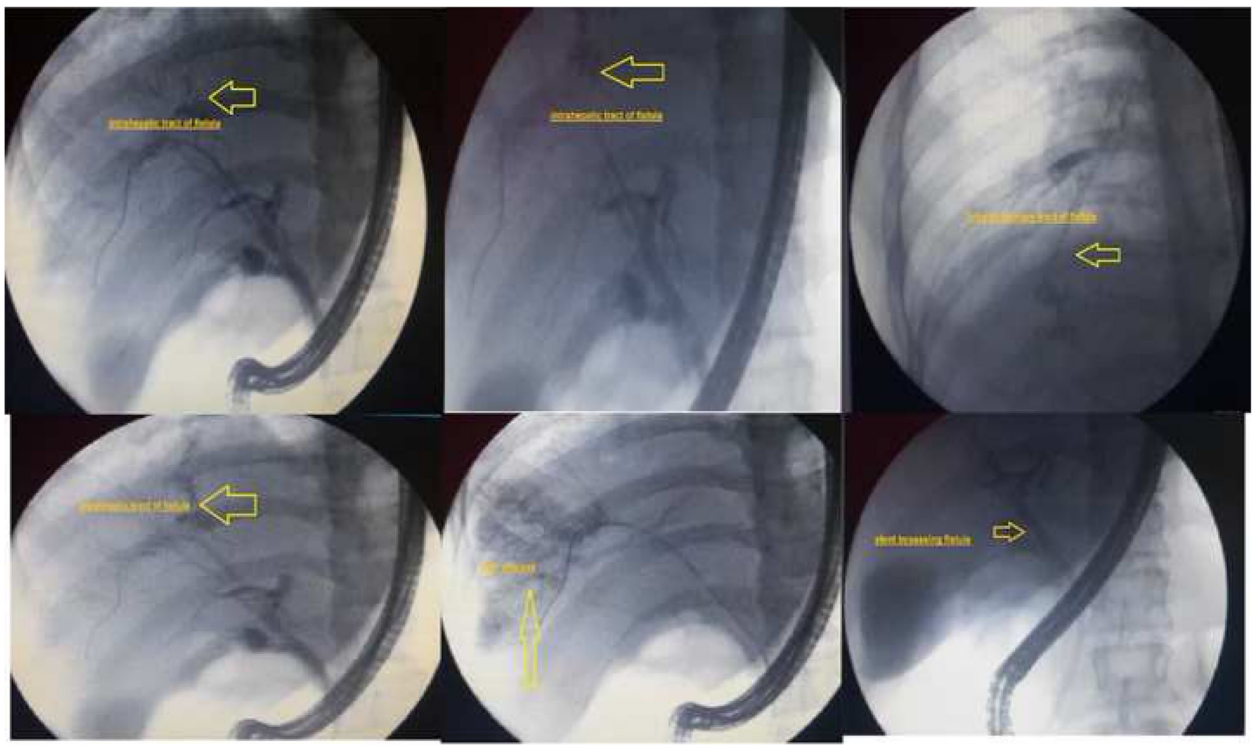

Fig. 2 ERCP fluoroscopic picture showing the abscess, the bronchobiliary fistula, and the biliary stent

Conclusion

ERCP stenting (endoscopic approach) is easy and is a possible management for cases of biliptysis.

Abbreviations

BBF: Bronchobiliary fistula; ERCP: Endoscopic retrograde cholangiopancreatography; PTD: Percutaneous transhepatic drainage

Acknowledgements

Thanks for my dear professor Dr. Gamal Badra Who gave me his experience in Endoscopy.

Author's contributions

OE write the whole case. The author read and approved the final manuscript.

Funding

No funding.

Availability of data and materials

The data and material are available.

Ethics approval and consent to participate

This study was approved by our institutional review board (IRB) (IRB00003413).

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Competing interests

The author declares that he has no competing interests.

Received: 1 August 2019 Accepted: 16 September 2019
Published online: 28 October 2019

References

1. SS AL-M, HH AL-J (1999) Chronic cough due to bronchobiliary fistula. Respiration 66(5):473–476. https://doi.org/10.1159/000029415
2. Liao GQ, Wang H, Zhu GY, Zhu K Bin, 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(33):3842–3849. doi:https://doi.org/10.3748/wjg.v17.i33.3842
3. Ragozzino A, De Rosa R, Galdiero R, Maio A, Manes G (2005) Bronchobiliary fistula evaluated with magnetic resonance imaging. Acta Radiol 46(5):452–454 http://www.ncbi.nlm.nih.gov/pubmed/16224917
4. Mehrzad H, Aziz A, Mangat K (2012) Transhepatic embolisation of a traumatic broncho-biliary fistula: a novel approach. Case Reports 2012(nov27 1): bcr2012006702–bcr2012006702. https://doi.org/10.1136/bcr-2012-006702
5. Yilmaz U, Sahin B, Hilmioglu F, Tezel A, Boyacioglu S, Cumhur T (1996) Endoscopic treatment of bronchobiliary fistula: report on 11 cases. Hepatogastroenterology 43(7):293–300 http://www.ncbi.nlm.nih.gov/pubmed/8682482
6. Fröbe M, Kullmann F, Schölmerich J, Böhme T, Müller-Ladner U (2004) Bronchobiliäre Fistel bei kombiniertem Lungen-Leber-Abszess. Med Klin 99(7):391–395. https://doi.org/10.1007/s00063-004-1057-4
7. Shim JR, Han S-S, Park HM, Lee EC, Park S-J, Park J-W (2018) Two cases of bronchobiliary fistula: case report. Ann Hepato-Biliary-Pancreatic Surg 22(2): 169. https://doi.org/10.14701/ahbps.2018.22.2.169
8. Ong M, Moozar K, Cohen LB (2004) Octreotide in bronchobiliary fistula management. Ann Thorac Surg 78(4):1512–1513. https://doi.org/10.1016/j.athoracsur.2003.10.137
9. Karabulut N, Çakmak V, Kiter G (2010) Confident diagnosis of bronchobiliary fistula using contrast-enhanced magnetic resonance cholangiography. Korean J Radiol 11(4):493. https://doi.org/10.3348/kjr.2010.11.4.493

Publisher's Note