A 48-year-old white woman, who worked in agriculture, was admitted with a 4-month history of cough productive of bile-like yellow phlegm. She had been hospitalized several times previously for recurrent right-sided pneumonias. The patient had undergone surgical resection of echinococcal cysts within liver segments V, VI, and VIII, cholecystectomy, and reconstruction of the right hepatic duct damaged by parasitic cyst infiltration 6 years previously (Fig. 1).

External drains had been inserted into both hepatic ducts through the common bile duct under radiological guidance (Fig. 2).

At subsequent outpatient visits, leakage of bile from the drains had persisted, and 3 months later, a plastic stent had been inserted into each hepatic duct during endoscopic retrograde cholangiopancreatography (ERCP). The patient had been advised to attend again for monitoring within 3–4 months with the possibility of replacement stents being required. However, she had refused to continue undergoing endoscopic procedures, and the stents remained in place for the next 6 years.

At the time of readmission, abdominal and chest computed tomography (CT) showed inflammatory lesions and a fluid collection in the subdiaphragmatic region; it was impossible to pinpoint the exact location of the fistula at bronchoscopy.

A subsequent CT and ERCP demonstrated the fistula between the biliary system and the lower lobe of the right lung (Figs. 3–5).

After endoscopic removal of the occluded plastic biliary stents, the patient was treated with antibiotics, and an improvement in her general condition and regression of symptoms were observed. A follow-up thoracic CT showed no inflammatory lesion in the lung tissue (Fig. 6).

The patient was discharged home after 8 days in good general condition. There were no recurrent symptoms at follow-up 24 months later.

A bronchobiliary fistula (BBF), a communication of the biliary system with the bronchial tree, is an exceptionally rare condition with underlying factors that include hydatid disease, hepatobiliary surgery, hepatic trauma, and congenital malformation [1–4]. The mechanism of transdiaphragmatic extension in BBF remains controversial. BBF due to the migration of short-term intrahepatic biliary stents has been previously reported [5].

To our knowledge, BBF caused by long-term extrahepatic biliary stenting has not previously been described, but has clearly been shown by our case, which also
demonstrates that in particular cases, the condition can be successfully managed with a conservative endoscopic approach.

Endoscopy_UCTN_Code_CPL_1AK_2AI

Competing interests: None

H. Razak Hady1, A. Baniukiewicz2, M. Luba1, P. Rogalski2, A. Dabrowski2, J. Dadan1

1 First Department of General and Endocrinological Surgery, Medical University of Bialystok, Poland
2 Department of Gastroenterology and Internal Medicine, Medical University of Bialystok, Poland

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Endoscopy 2011; 43: E178 – E179
© Georg Thieme Verlag KG Stuttgart · New York · ISSN 0013-726X

Corresponding author
H. Razak Hady, MD
Medical University of Bialystok
ul. M. Sklodowskiej-Curie 24a
15-276 Bialystok
Poland
Fax: +48-85-7468620
khchirog@umwb.edu.pl