Case Report

Broncho-biliary fistula secondary to biliary obstruction and lung abscess in a patient with pancreatic neuro-endocrine tumor

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Abstract We present a case report of broncho-biliary fistula that developed due to the blockage of biliary stent placed during the management of pancreatic neuroendocrine tumor (pNET); diagnosed on high clinical suspicion, percutaneous cholangiogram and contrast enhanced computed tomography (CECT); and successfully treated with percutaneous transhepatic biliary drainage (PTBD).

Introduction

Broncho-biliary fistula (BBF) is defined as the passage of bile in the bronchi and the presence of bile in the sputum (biliptysis). BBF usually occurs either in the congenital form [1] or following thoraco-abdominal insult. BBF is a rare clinical presentation which was first reported in the year 1850 [2]. BBF has been commonly seen in hepatic malignancy [3,4]. Other causes of BBF include lung abscess [5], sub-phrenic abscess [6], postoperative stenosis (including biliary/gastric or pancreatic surgery [7]), amebic abscess [7,8], echinococcosis [9,10], pyogenic liver abscess [11] and traumatic/surgical injury [12–14] to hepatobiliary system resulting in the obstruction of bile flow. BBF has been reported as post-surgical complication in many malignancies like cholangiocarcinoma [4], gall bladder carcinoma [13] and hepatic metastases [14] due to colorectal carcinoma [4], but to the best of our knowledge this complication has never been reported in clinical or therapeutic course of neuroendocrine tumor of pancreas.

Case report

A 37 year old female was diagnosed as a case of metastatic pNET when she presented with a severe backache in August 2012. She had a past history of lateral pancreatico-jejunostomy for chronic pancreatitis in March 2012.

At the time of presentation, positron emission tomography–computed tomography (PET–CT) scan revealed an enhanced lesion in the region of the head and uncinate process.
of the pancreas with a few enlarged lymph nodes (LN) along with multiple skeletal metastases. There was dilatation of middle and proximal common bile duct (CBD) with upstream bilobar intrahepatic biliary radical dilatation (IHBRD). She underwent endoscopic retrograde cholangio-pancreatography (ERCP) guided pancreatico-biliary stenting for obstructive jaundice. Fine needle aspiration cytology (FNAC) from peripancreatic LN in September 2012 showed neuroendocrine tumor cells that were positive for synaptophysin and chromogranin-A. Ki 67 index was 4-5%, indicating a low grade tumor.

She received palliative radiotherapy and zoledronic acid for painful skeletal metastases. Patient received everolimus and octreotide therapy for six months. After progression on above therapy patient received peptide receptor radiotherapy (PRRT), four sessions till October 2014.

In January 2015 she presented with the complaints of fever and cough for one month. Radiological evaluation revealed left sided loculated empyema with concomitant left sided lung abscess (Fig. 1). Intercostal drainage tube was placed for the drainage of loculated empyema (Fig. 2) along with single aspiration of lung abscess. Microbial culture of lung abscess showed the growth of *Escherichia coli* (*E. coli*). Antibiotics were started accordingly. Although fever subsided cough aggravated with the production of green colored sputum (more of serous in consistency) and patient was unable to lie down due to aggravation of cough. High resolution computed tomography (HRCT) of chest showed a thick walled cavitary lesion in the antero-lateral-basal segment of the lower left lobe with loss of fat planes with the diaphragm.

Contrast enhanced computed tomography (CECT) of the abdomen (Fig. 3) revealed dilated intrahepatic biliary radicles with heterogeneous enhancement in segment II of the liver. There was a loss of fat planes between the involved hepatic segment, adjacent left dome of diaphragm and the basal segment of lung suggestive of possible contiguity.

Percutaneous cholangiogram (Fig. 4) showed biliary ductal dilatation and subtle contrast leak from left hepatic duct with pooling in adjoining area (left lung base) along with blockage of previously placed biliary stent.

In view of the above findings and increasing discharge of green colored sputum of 400 ml/day, bilipysis due to broncho-biliary fistula was suspected. Percutaneous transhepatic biliary dilatation (PTBD), patient showed a marked improvement in symptoms. Bilipysis stopped immediately following PTBD procedure. At one week follow up, ERCP cholangiogram was done which did not show broncho-biliary fistula, indicating spontaneous sealing of fistula. CBD stent was replaced. PTBD tube was removed after 48 h of ERCP placed.
CBD stent. Patient’s general condition improved with no fever, cough or expectoration.

Discussion

The most common clinical symptom of BBF is bilipysis, which is seen in almost 100% patients [3]. Other symptoms include respiratory symptoms, such as irritating cough, fever and jaundice.

The diagnosis of BBF can be done by different modalities like percutaneous cholangiogram [9], magnetic resonance cholangio-pancreatography (MRCP) [15], 99mTcMebrofeninscintigraphy [9], hepatobiliary-imino-diacetic acid (HIDA) scan [4], cholescintigraphy with single-photon emission computed tomography (SPECT), sputum examination for bilirubin, CECT study [12] and bronchoscopy [4,5]. In our case the diagnosis was made using CT scan and cholangiogram.

Treatment modalities for BBF may include a more aggressive approach like surgery i.e. excision of the broncho-biliary fistula. Less-invasive and commonly used approach with which most of the patients can be treated includes PTBD [8], ERCP with sphincterotomy and stenting [12], interventional transhepatic embolization with microcoils and Endobronchial embolization using silicone spigots [4].

In our case green colored sputum (more of serous in consistency) raised a high clinical suspicion of bilipysis. Diagnosis of BBF was confirmed by CECT and percutaneous cholangiogram. Decompression of the obstructed biliary system was done by PTBD followed by ERCP and stent replacement.

The expected mechanism for the development of BBF in our case is most likely due to biliary obstruction caused by progressive neuroendocrine tumor of pancreatic head and blockage of CBD stent. This was further complicated by biliary tract infection causing the formation of cholangiolar abscess with contiguous involvement of diaphragm and lung thereafter. The presence of \textit{E. coli}, an enteric gram negative bacteria in the pleural fluid points to this fact.

In view of excellent response to this therapy, we may make an opinion that in a clinical context the level of suspicion should be high and timely diagnosis can let clinicians help in exploring non surgical treatment modalities, which can deliver desirable results.

Conflict of interest

The author(s) declare(s) that there is no conflict of interest regarding the publication of this paper.
Figure 3  Coronal reformatted contrast enhanced CT image showing heterogeneous enhancement in segment II of liver. There is a loss of fat plane between the involved hepatic segment, adjacent left dome of diaphragm and the basal segment of lung suggestive of possible contiguity (arrow).

Figure 4  Percutaneous cholangiogram showing biliary ductal dilatation and subtle contrast leak from left hepatic duct with pooling in adjoining area (left lung base).
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