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Case Report

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Special Presentation of Bronchobiliary Fistula After Transcatheter Arterial Chemoembolization: A Case Report

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Abbreviations:

TACE: Transcatheter arterial chemoembolization; HCC: Hepatocellular carcinoma; BCLC: Barcelona Clinic Liver Cancer; BBF: Bronchobiliary fistula

1. Abstract

Transcatheter Arterial Chemoembolization (TACE) has been widely adopted for Hepatocellular Carcinoma (HCC) treatment. However, it may cause several complications, including biloma formation. And bronchobiliary fistula is a post-TACE complication rarely mentioned before.

Herein we share a 65-year-old man encountering fever, dyspnea, abdominal pain, and abundant yellowish purulent bronchorrhea after TACE procedure. CT scan of his chest showed a connection between the right lower lobe of the lung and the hepatic cystic lesion. We placed a percutaneous drain and obtained yellowish bile with the same characteristics as the patient's bronchorrhea. Unfortunately, a metastatic brain tumor was diagnosed. Two weeks later, the patient passed away due to progressive right lower lobe pneumonia.

This case exhibits a clinical presentation of post-TACE bronchobiliary fistula and typical CT scan image which was helpful for diagnosis. Biliary tree injuries with subsequent biloma formation and diaphragmatic injuries may contribute to the formation of bronchobiliary fistula. Therefore, we put forward this rare complication in hopes that future clinicians will keep earlier intervention in mind when post-TACE bronchobiliary fistula occurs.

2. Introduction

Liver cancer is a challenging worldwide disease with an estimated incidence of >1 million cases by 2025 [1]. Hepatocellular Carcinoma (HCC) is the most common type of liver cancer and is highly related to the infection of hepatitis B virus and hepatitis C viruses. Transcatheter Arterial Chemoembolization (TACE) has been widely adopted for patients with intermediate-stage HCC, who are not suitable for resection [1]. Despite its significant anti-tumor effects, the TACE procedure may cause hepatic and biliary damage, liver abscess, and intrahepatic biloma formation [2]. Conservative percutaneous drainage can manage biloma in most cases and have a good prognosis. In this case report, we share a rare complication, bronchobiliary fistula, related to TACE-induced biloma.

3. Case Report

This 65-year-old man with a history of chronic hepatitis C was diagnosed with Hepatocellular Carcinoma (HCC), cT4N1M0, Barcelona Clinic Liver Cancer (BCLC) stage C. He underwent six cycles of TACE and received two courses of palliative radiation therapy for HCC in the S4 and S6 of the liver in the past 15 months. He also took Sorafenib (Nexavar®). Computed Tomography (CT) scan three months before this admission showed a cystic lesion with surround-

ing daughter cysts without ring enhancement developed in S7, where the previous HCC was located, so biloma was highly suspected. A week before this admission, he encountered sudden onset of fever, dyspnea, and yellowish sputum for a day. Thus, he sought medical help at another hospital and was admitted with tentative diagnoses of liver abscess and pneumonia. He underwent percutaneous abscess drainage, and they prescribed piperacillin/tazobactam and meropenem for five days. However, there were no significant improvements, except for his fever. The bacterial culture from drainage revealed Citrobacter koseri. Subsequently, the patient was transferred to our hospital.

At our hospital, he presented symptoms of severe right upper quadrant abdominal pain (Visual Analogue Scale:5-6), jaundice, dyspnea, abundant yellowish purulent bronchorrhea, and yellowish nasal discharge (Figure 1). On examinations, there were decreased breath sounds with rales in the right lower lung. The hemogram showed leukocytosis with the left shift. Blood levels of hsCRP and total bilirubin were 19.42 mg/dL and 2.33 mg/dL, respectively. There was no leukocyte or bacteria in his sputum culture. His chest CT scan showed a connection between the right lower lobe of the lung and the hepatic cystic lesion (Figure 2). We placed an 8 Fr ring catheter percutaneous drainage for the suspected biloma and obtained yellowish bile with the same characteristics as the patient's bronchorrhea. According to the CT findings and the presentation of cough with bilioptysis, we confirmed the diagnosis of biloma with bronchobiliary fistula. The followed-up CT scan four weeks later showed shrinkage of biloma. Because of persisted bilioptysis, we recommended he undergo surgical ligation for the curative treatment of BBF. Unfortunately, left hemianopsia developed gradually, and the brain magnetic resonance imaging (MRI) showed a metastatic tumor in the right occipital region. Furthermore, his dyspnea with desaturation progressed, so we cancelled surgical intervention. Two weeks later, the patient passed away due to progressive right lower lobe pneumonia.

Figure 1: Bilioptysis developed during the hospital course

Figure 2: CT scan revealed a diaphragmatic defect (arrow) and a connection between biloma and bronchial cyst with an air-fluid level. In addition, an inflammatory pulmonary consolidation in the lateral basal segment of the right lower lobe was also visible

4. Discussion

Biloma is a complication of transcatheter arterial chemoembolization (TACE). Previous studies found the incidence of intrahepatic biloma was 1.04% after TACE [2]. According to the past literature, the mean interval time was 69.1 days between intrahepatic biloma formation and the most recent TACE procedure [2]. Bronchobiliary Fistula (BBF), defined as an abnormal channel between the biliary system and bronchial tree through the diaphragm, is rare [3, 4]. Bronchobiliary fistula formed after TACE-induced biloma had been reported even less; its prevalence is currently unknown. We hope to highlight the unique presentation of bronchobiliary fistula formation after TACE-induced biloma progressed.

A diagnosis of bronchobiliary fistula is made mainly based on clinical symptoms and imaging [3]. Manifestations include bilioptysis, pleural effusion, atelectasis, liver abscess (cyst), and intrahepatic bile duct dilatation [3, 5]. Previous reports concluded that the fistulous tract connecting pleural effusion and biliary lesions is rarely seen on CT, namely 2 out of 11 patients [3]. In our case, however, the diaphragmatic defect was visibly notable on the CT scan. Furthermore, there was an air-fluid level in the cyst, making it clear that a connection between the biloma and bronchus existed. These all served as distinguishable features on CT for bronchobiliary fistula.

Although the etiology of BBF formation after TACE remains unclear, a few reasons can be considered. First of all, injuries to the biliary tree. TACE procedure may cause the peribiliary capillary plexus to fill with iodized oil or other embolic materials [2]. Therefore, ischemia of the intrahepatic bile ducts may easily occur and lead to biloma formation [2, 6]. Bilomas can then cause an increase in intra-

biliary or intracavitary pressure, which possibly drives bile into the chest [3]. Diaphragmatic injuries worsen this condition and might be the precipitating factor. Besides, infection, thermal injury by radiofrequency ablation, and surgical damage of the diaphragm can all contribute to the development of BBF [3, 7, 8]. As in our case, even though the TACE procedure did not directly injure the diaphragm, bacteria isolated from the biloma may contribute to diaphragmatic injuries and form a bronchobiliary fistula.

Most BBF can be managed successfully by drainage or close monitoring [4]. However, ongoing bile leaks may require further management, such as surgical fixation [3, 5, 9, 10]. There is no official guideline for the treatment of bronchobiliary fistula. A systematic literature review suggests open surgery should be the first choice after interventional techniques have failed or when BBF is secondary to tumors and biliary obstruction [5]. In our case, the patient's condition progressed after he underwent the intervention of percutaneous drainage. Therefore, we consulted a surgeon for fistula ligation. However, his dyspnea with desaturation developed later, so we cancelled surgical intervention.

In conclusion, a small proportion of patients might form biloma after TACE therapy and may subsequently cause BBF, especially those present with infection. CT scan can be a way to confirm the diagnosis of BBF. This case demonstrates that we should be aware of the possibility of BBF formation after the TACE procedure, which can be a fatal complication. Moreover, patients may benefit from earlier surgical intervention.

References

1. Llovet JM, Kelley RK, Villanueva A, Singal AG, Pikarsky E, Roayaie S, et al. Hepatocellular carcinoma. Nature Reviews Disease Primers. 2021; 7(1): 6.
2. Zhang B, Guo Y, Wu K, Shan H. Intrahepatic biloma following transcatheter arterial chemoembolization for hepatocellular carcinoma: Incidence, imaging features and management. Mol Clin Oncol. 2017; 6(6): 937-43.
3. Huang Z-M, Zuo M-X, Gu Y-K, Lai C-X, Pan Q-X, Yi X-C, et al. Bronchobiliary fistula after ablation of hepatocellular carcinoma adjacent to the diaphragm: Case report and literature review. Thorac Cancer. 2020; 11(5): 1233-8.
4. Varela Vega M, Durán F, Geribaldi N, San Martín G, Ettlin A. Hepatobronchial Fistula: A Rare Complication of Liver Abscess. Cirugía Española (English Edition). 2017; 95(7): 410-1.
5. Liao G-Q, Wang H, Zhu G-Y, Zhu K-B, Lv F-X, Tai S. Management of acquired bronchobiliary fistula: A systematic literature review of 68 cases published in 30 years. World J Gastroenterol. 2011; 17(33): 3842-9.
6. Kobayashi S, Nakanuma Y, Terada T, Matsui O. Postmortem survey of bile duct necrosis and biloma in hepatocellular carcinoma after transcatheter arterial chemoembolization therapy: relevance to microvascular damages of peribiliary capillary plexus. Am J Gastroenterol. 1993; 88(9): 1410-5.
7. Shim JR, Han S-S, Park HM, Lee EC, Park S-J, Park J-W. Two cases of bronchobiliary fistula: Case report. Ann Hepatobiliary Pancreat Surg. 2018; 22(2): 169-72.
8. Acquasanta M, Spadarella G, Civelli EM. Spontaneous bronchobiliary fistula: Case report. J Radiol Case Rep. 2020; 14(10): 10-5.
9. Copelan A, Bahoura L, Tardy F, Kirsch M, Sokhandon F, Kapoor B. Etiology, Diagnosis, and Management of Bilomas: A Current Update. Tech Vasc Interv Radiol. 2015; 18(4): 236-43.
10. Eryigit H, Oztas S, Urek S, Olgac G, Kurutepe M, Kutlu CA. Management of acquired bronchobiliary fistula: 3 case reports and a literature review. J Cardiothorac Surg. 2007; 2: 52.