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Case Report

Synchronous occurrence of pulmonary sarcomatoid carcinoma and intrahepatic cholangiocarcinoma: a case report and literature review

Jia-Jin Yang, Yan-Jun Lu, Jian-Bing Wu

Department of Oncology, The Second Affiliated Hospital of Nanchang University, Nanchang 330006, China

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Abstract: Pulmonary sarcomatoid carcinoma (PSC) is a rare histologic subtype of non-small cell lung cancer (NSCLC). The incidence ranges from 0.1% to 4.7% of all lung malignancies. However, Synchronous Intrahepatic cholangiocarcinoma (ICC) with primary pulmonary sarcomatiod carcinoma are much rarer double-cancer. Such a case has not previously been reported till now according to the literature. The coinciding malignancy usually been discovered during surgical exploration. Thus, this is the first report where a targeted biopsy of the clinically suspicious lesion was used to determine the diagnosis of pulmonary sarcomatiod carcinoma concurrent with intrahepatic cholangiocarcinoma. Herein, we present such a rare case and review of the literature.

Keywords: Sarcomatoid carcinoma, lung, non-small cell lung cancer, intrahepatic cholangiocarcinoma

Introdution

Pulmonary sarcomatoid carcinoma (PSC) is a rare histologic subtype of non-small cell lung cancer (NSCLC), which be defined as poorly differentiated non-small cell carcinoma that contains a component of sarcoma or sarcoma-like with giant and/or spindle cells according to the 2004 World Health Organization (WHO) [1]. The incidence ranges from 0.1% to 4.7% of all lung malignancies [2]. However, synchronous primary PSC with intrahepatic cholangiocarcinoma (ICC) is a rarer double-cancer. According to the literature, such a case has not previously been reported till now, and no previous cases have been diagnosis by the targeted biopsy of a clinically suspicious lesion. The current study reports the rare case of targeted biophy-identified PSC concurrent with ICC.

Case presentation

On June 15, 2015, a 61-year-old man was admitted to our department because of abdominal discomfort and weight loss for half month. He had a 30-year history of cigarette smoking (15 cigarettes per day). He denied alcohol use, any occupational exposures, allergies, and any significant medical history before. Laboratory studies showed a slightly elevated C-reactive protein of 32.9 mg/L (0.0 to 8.0 mg/L), leukocytosis of 12.89×10^9/L (4 to 10×10^9/L), a normal renal and liver function and showed no evidence of past or persistent hepatitis B virus or hepatitis C virus infection. The tumor markers of carbohydrate antigen (CA) 19-9 showed a slightly elevation, while other markers such as α-fetoprotein (AFP), CA 125 and carcinoembryonic antigen (CEA) were all within the normal ranges. Contrast-enhanced computed tomography (CT) of the abdomen and chest showed a hypodense hepatic lesion consisting of a 5.7×6.8 cm mass with irregular margins in the left lobe of the liver, and showed peripheral rim enhancement in the arterial phase, and progressive hyperattenuation on venous and delayed phase (**Figure 1B**). Various enlarged lymph nodes were observed in the porta hepatis and retroperitoneal space. Chest CT showed an ovoid lesion about 3.3 cm in diameter in the middle lobe of the right lung with enlarged lymph nodes in the mediastinal and armpit (**Figure 1A**). Bone scan and electronic.

Gastroscope showed no abnormality. For definite diagnosis of the disease, percutaneous lung and liver biopsy under CT were performed

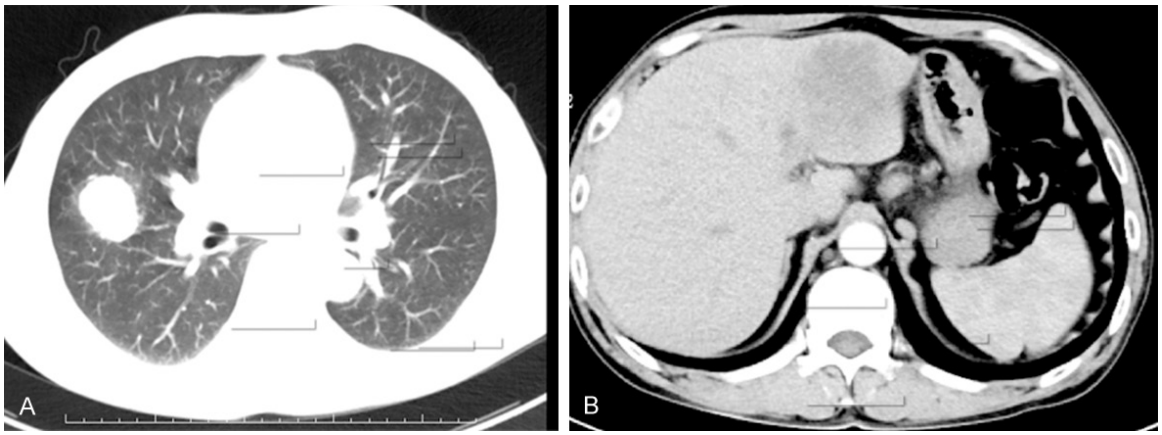

Figure 1. Chest and abdominal Contrast-enhancced computed tomography of the patient. A. Chest CT showed an ovoid lesion in the middle lobe of the right lung. B. Abdominal CT showed a hypodense hepatic lession with irregular margins in the left lobe of the liver, and showed peripheral rim enhancement in the arterial phase.

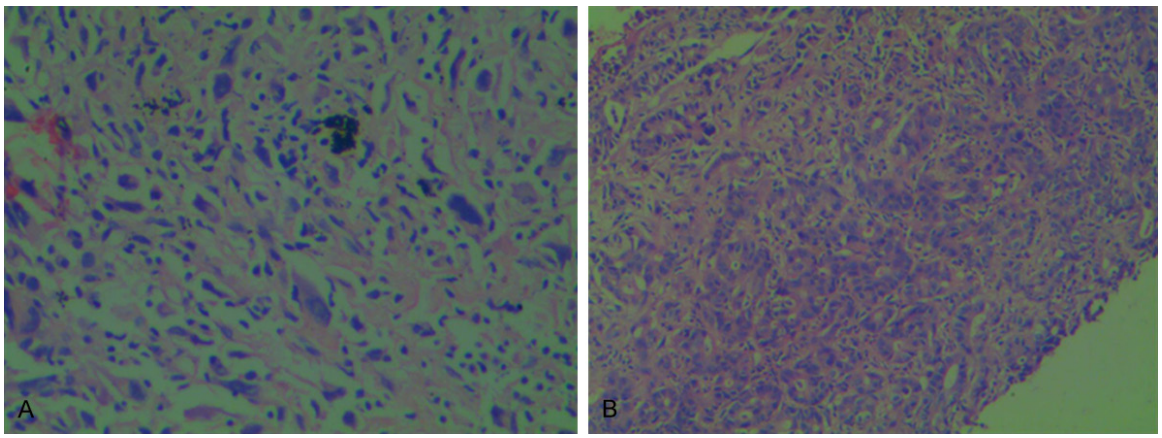

Figure 2. Histological examinationof the percutaneous lung and liver biopsy. A. Lung specimen showed Tumor cells were pleomorphic, and spinder-like in shape (Hematoxylin and eosin staining; magnification, ×200). B. Liver specimen showed malignant cells with a glandular structure, which was consistent with adenocarcinomal (Hematoxylin and eosin staining; magnification, ×100).

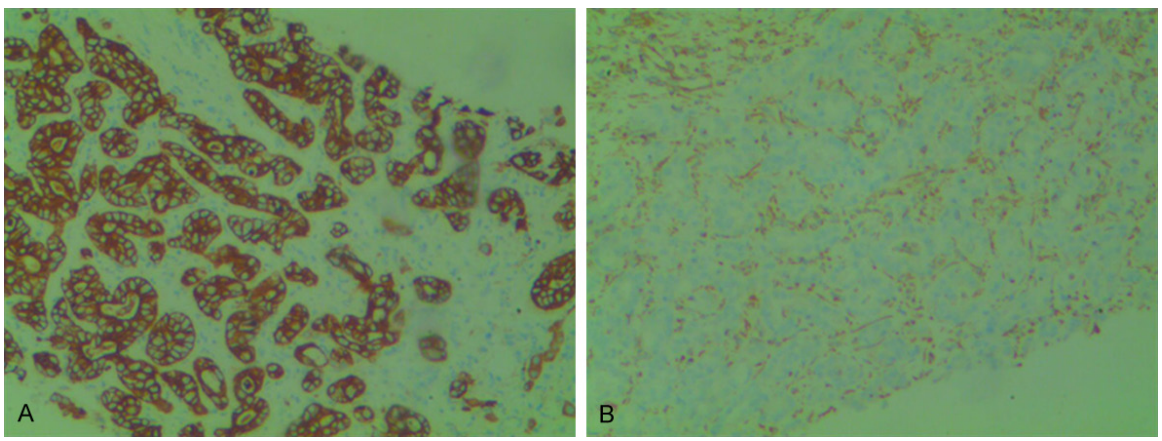

Figure 3. Immunohistochemical staining demonsrates that tumor cells of lung specimen were (A) positive for CK and (B) positive for vimentin.

in our department. Histological examination of the lung specimen showed pleomorphic, and spindle cells (**Figure 2A**). Immunohistochemically, the tumor cells were positive for cytokeratin (CK) (**Figure 3A**), CK19, Vimentin (**Figure 3B**), and thyroid transcription factor-1 (TTF-1), while they are negative for CK7, S100, and CEA. The morpholoy and immunohistochemistry are most consistent with sarcomatoid carcinoma NSCLC. Histological examination of the liver specimen showed malignant cells with a glandular structure, which was consistent with adenocarcinomal (**Figure 2B**).

Immunohistochemically, the tumor cells were positive for CK, CK19, and Villin, while negative for Vimentin, TTF-1, CK7, CK20, and caudal-related homeobox 2 (CDX2). The final diagnosis was synchronous PSC and ICC.

Discussion

Synchronous primary PSC with ICC is much rarer double-cancer, which has not been reported till now according to the literature. Smoking is the most possible risk factors to the double-cancer, for PSC and ICC have the consistent risk factors of smoking [3-6]. The patient had a 30-year history of cigarette smoking. Kai Mao et al. [7] have reported that lung cancer could be increased the standardized incidence ratios (1.75) of cholangiocarcinoma. The increased risk may be due to shared genetic or environmental etiological factors between PSC and ICC.

The liver is one of the most common metastatic sites of NSCLC, with incidence rate of 38 to 44% at previous study. Lung metastasis can also happen in liver cancer. So the diagnosis of this disease is challenging, and the coinciding malignancy usually been discovered during surgical exploration. We have performed percutaneous lung and liver biopsy under CT to diagnose this double-cancer. The diagnosis of PSC which consist of epithelial and sarcomatous/sarcomatoid component, mainly depends on morphologic characteristics and immunohistochemical staining. The epithelial component is stained with antibodies to cytokeratins, epithelial membrane antigen (EMA), and carcinoembryonic antigen (CEA), whereas the opposite holds true for the sarcomatous or sarcomatoid component, which is instead consistently immunoreactive for vimentin [8, 9]. Pelosi et al. [10] reported that vimentin could be an effective tool to reliably distinguish PSC from other NSCLC in small biopsy specimens. So their confirm that a strong and diffuse vimentin expression by immunohistochemical by sarcoma-like tumor cells featuring spindle and/or giant cell changes would authorize diagnosis of PSC in biopsy samples. In our case, we found that the PSC specimen was positive for CK, TTF-1 and vimentin, but negative for CEA, CK7 and S100, which could be diagnosed of PSC. In our case, histologic examination of the liver specimen was consistent with adenocarcinomal, which have a difference cellular-origin of PSC. We have to identify whether the liver tumor metastatic from gastrointestinal malignant cell. However, electronic gastroscope in our case showed no abnormality. The histological feature of our case was similar to the previous study [11]. So, the final diagnosis was synchronous PSC and ICC.

In conclusion, a rare case of synchronous primary PSC and ICC is presented, which was diagnosed by a targeted biopsy of the suspected lesion. The diagnosis of this disease mainly depends on morphologic characteristics and immunohistochemical staining. Such a case has not previously been reported till now according to the literature. Multicenter collaborations are necessary to determine the optimal treatment.

Disclosure of conflict of interest

None.

Address correspondence to: Jia-Jin Yang, Yan-Jun Lu and Jian-Bing Wu, Department of Oncology, The Second Affiliated Hospital of Nanchang University, Nanchang 330006, China. E-mail: y6609711@163.com (JJY); ianlyj@163.com (YJL); 15270880537@163.com (JBW)

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