Hemothorax caused by spontaneous rupture of a metastatic mediastinal lymph node in hepatocellular carcinoma: a case report

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To the Editor,
The frequency of massive hemoperitoneum caused by spontaneous rupture of hepatocellular carcinoma (HCC) has been reported to be 10% to 18% because of the extensive vascular structure and relatively small amount of fibrous tissue in these tumors [1]. Hemothorax caused by rupture of a lung or pleural metastasis of HCC occurs less frequently. Although one case of cardiac tamponade caused by spontaneous rupture of a metastatic mediastinal lymph node (MLN) has been described [2], to our knowledge, no case of hemothorax due to spontaneous rupture of a metastatic MLN of HCC has been reported in the Korean- or English-language literature.

We describe here a case of massive hemothorax due to spontaneous rupture of a metastatic MLN in HCC. A 60-year-old male Korean farmer was brought to the Department of Emergency, Ulsan University Hospital with dyspnea and left pleuritic chest pain within 6 hours after symptom onset.

Six years earlier, he was diagnosed with chronic hepatitis B-related cirrhosis and 4 years earlier he had been diagnosed with HCC (Fig. 1A) and underwent a right hepatic lobectomy. Multiple metastatic pulmonary nodules were detected 3 months after the surgery and the patient received six cycles of a cisplatin-based chemotherapy regimen over 6 months. Contrast-enhanced computed tomography (CT) scanning showed complete disappearance of the multiple metastatic lung nodules after chemotherapy. At 8 months after finishing the chemotherapy, however, he was readmitted to our hospital due to a single metastatic nodule in the left lower lobe of the lung and underwent metastasectomy with video-assisted thoracoscopic surgery (VATS). Pathological examination of the lung nodule removed showed results consistent with metastatic HCC.

At 1 year before admission, CT images of the chest showed a single enlarged lymph node (LN) in the left inferior pulmonary ligament, regarded as a metastatic MLN of HCC (Fig. 1B). Two weeks before this admission, follow-up CT images showed enlargement of the metastatic MLN, and the patient was scheduled for additional chemotherapy.

On the day of admission, the patient experienced an abrupt onset of dyspnea and left pleuritic chest pain. Physical examination on admission revealed an acutely ill-looking man with body temperature of 36.5°C, pulse rate of 130 beats per minute,
blood pressure of 100/70 mmHg, and a respiration rate of 32 breaths per minute. Laboratory test results included hemoglobin 12.3 g/dL, hematocrit 33.4%, white blood cell count $4.37 \times 10^3/\mu$L, platelet count $9.9 \times 10^4/\mu$L, aspartate aminotransferase 47 IU/L, alanine aminotransferase 60 IU/L, total bilirubin 2.0 mg/dL, albumin 3.2 mg/dL, and $\alpha$-fetoprotein 819.2 ng/mL. Chest X-rays showed a massive left-sided pleural effusion, with the trachea deviated to the right side.

Massive hemothorax was diagnosed by thoracentesis. CT images of the chest revealed a $73 \times 84$ mm-sized ruptured low-attenuated central necrotic mass with massive left side hemothorax (Fig. 1C). A chest tube was inserted and approximately 1,200 mL of bloody

**Figure 1.** Chest computed tomography scans showing (A) a 10-cm sized intrahepatic peripheral capsular enhanced mass, (B) a single enlarged lymph node (white arrow) in the left inferior pulmonary ligament, and (C) a $73 \times 84$-mm ruptured low-attenuated central necrotic mass with a large amount of pleural fluid in the left hemithorax. (D) Chest X-ray showing bronchial arterial angiography after injection of lipiodol ultrafluide and adriamycin (doxorubicin hydrochloride) with polyvinyl alcohol particles (contour emboli).
fluid was drained. Arteriography of the bronchial arteries revealed a massive ruptured LN in the left inferior pulmonary ligament, to which blood was supplied by an accessory bronchial artery originating 10 cm beneath the left bronchial artery.

These findings indicated that the massive hemothorax was caused by a spontaneous rupture of a metastatic MLN of HCC into the intrapleural space. Transcatheter arterial embolization (TAE) of the left bronchial artery was performed successfully, injecting 13 mL of lipiodol ultrafluide (Guerbet, Aulnay-sous-Bois, France) and 30 mg of adriamycin (doxorubicin hydrochloride) with polyvinyl alcohol particles (contour emboli, Interventional Therapeutics Corp., Fremont, CA, USA) (Fig. 1D). No complication was observed and the pleural effusion gradually disappeared thereafter.

The patient’s dyspnea improved, as did his physical condition. Following removal of the chest tube, he was discharged after 15 days in the hospital and was followed monthly as an outpatient at the Department of Oncology for 3 months. Serial chest X-rays revealed a decrease in the size of the MLN with lipiodol embolization. Four months later, however, the patient died at home.

Due to its vascular structure and relatively small amount of fibrous tissue, spontaneous rupture of HCC is not uncommon [1]. Rupture of HCC is considered a medical emergency and is associated with high mortality. HCC frequently metastasizes, most often to the lungs, LNs, bones, and adrenal glands. Moreover, HCC metastases, like the primary tumors, may rupture spontaneously.

Sohara et al. [3] reviewed 10 cases with HCC complicated by hemothorax, including four case reports in Japanese, describing patients with metastasis to the chest wall and rib, lung, pleura, diaphragm, and MLN [4]. Those reports included the first case of hemothorax from spontaneous rupture of a mediastinal metastasis [4].

Common clinical presentations are chest pain and dyspnea initially [3]. Other signs are palpitations and hypotension, consistent with hypovolemic shock. Reported rare signs included massive hemoptysis and respiratory failure. Our patient also developed hemothorax with sudden-onset chest pain, dyspnea, and tachycardia. Ruptured HCC can be treated surgically or by TAE, with the latter now used widely for HCC ruptured into the peritoneal cavity. Masumoto et al. [5] first reported hemothorax due to HCC rupture successfully controlled by TAE and our case was also successfully controlled using TAE. On the other hand, surgically treated and untreated failures have been reported and drainage-only cases do allow complete control [3].

Our patient had been diagnosed with HCC 4 years earlier and had undergone various treatments, including right hepatic lobectomy, six cycles of chemotherapy for multiple lung metastases, VATS metastasectomy for a single metastasis in the lung, and TAE for rupture of metastatic MLN causing massive hemothorax. TAE was effective, in that bleeding was successfully controlled and pleural effusion did not recur. Our findings suggest that patients with HCC should be closely monitored and suitably managed to improve survival.

In conclusion, this is the first report of hemothorax secondary to spontaneous rupture of metastatic MLN of HCC in the Korean- or English-language literature. The hemothorax was successfully treated with TAE. The various manifestations observed in patients with HCC suggest the need for careful monitoring and suitable management.

Keywords: Carcinoma, hepatocellular; Neoplasm metastasis; Hemothorax

Conflict of interest
No potential conflict of interest relevant to this article is reported.

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