



Case report

Pulmonary lymphangitic carcinomatosis secondary to ureteral cancer



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ABSTRACT

Background: Pulmonary lymphangitic carcinomatosis (PLC) is a metastatic lung disease of malignant tumors that spread through pulmonary lymphatic vessels. Although prompt diagnosis and specific treatment of PLC are required due to the poor prognosis associated with this disease, it is often challenging to determine the primary cancer site.

Case presentation: A 67-year-old Japanese woman presented to our hospital with a 10-day history of cough and dyspnea on exertion. Chest radiography and computed tomography (CT) revealed diffuse nodular opacities with interlobular septal thickening. Both bronchoalveolar lavage (BAL) and transbronchial lung biopsy (TBLB) revealed carcinoma cells with unknown origin. Contrast-enhanced CT depicted a mass in the right ureter with hydronephrosis, and retrograde urography showed a narrowing of the right ureter. Urine cytology from her right ureter via ureteral catheter also revealed atypical cells, highly suggestive of malignancy. Immunohistochemical examination of lung specimens via TBLB showed results consistent with lung metastasis of ureteral cancer. Therefore, we arrived at a diagnosis of PLC secondary to ureteral cancer.

Conclusions: This case encouraged multidisciplinary discussion and a whole-body examination, including TBLB with immunohistochemistry, to determine the origin of PLC.

1. Background

Pulmonary lymphangitic carcinomatosis (PLC) is a metastatic lung disease of malignant tumors that spread through pulmonary lymphatic vessels. Although prompt diagnosis and specific treatment of PLC are required due to the poor prognosis associated with this disease, it is often challenging to determine the primary cancer site. This is the case report of PLC secondary to ureteral cancer.

2. Case presentation

A 67-year-old Japanese woman with 10 pack-years of cigarette smoking presented to our hospital with a 10-day history of cough and dyspnea on exertion. All of the vital signs were within normal range except for peripheral oxygen saturation of 93% on room air. The physical examination revealed coarse crackles in the lungs bilaterally on auscultation although there was no heart murmur.

Chest radiography revealed bilateral reticular opacities and bilateral

blunted costophrenic angles (Fig. 1). Furthermore, computed tomography (CT) showed diffuse nodular opacities with interlobular septal thickening and bilateral pleural effusions without suspicion of primary lung cancer (Fig. 2). The serum levels of carcinoembryonic antigen (CEA) and cytokeratin 19 fragment (CYFRA) were elevated at 24.9 ng/mL and 14.4 ng/mL, respectively. Her serum B-type natriuretic peptide was 8.3 pg/mL; thus, heart failure was unlikely. These radiologic and laboratory findings strongly suggested PLC. We considered gastric cancer as the primary tumor and performed an upper gastrointestinal endoscopy, however, there were no abnormalities detected. A colonoscopy could not be performed because of her poor general condition.

Both bronchoalveolar lavage (BAL) and transbronchial lung biopsy (TBLB) revealed carcinoma cells with unknown origin. Furthermore, contrast-enhanced CT depicted a mass in the right ureter with hydronephrosis. Urinalysis revealed no occult bleeding. Retrograde urography showed a narrowing of the right ureter (Fig. 3), and urine cytology from the right ureter via ureteral catheterization also found atypical cells, highly indicative of malignancy. Immunohistochemistry of lung

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Fig. 1. Chest radiography shows bilateral reticular opacities and bilateral blunted costophrenic angles.

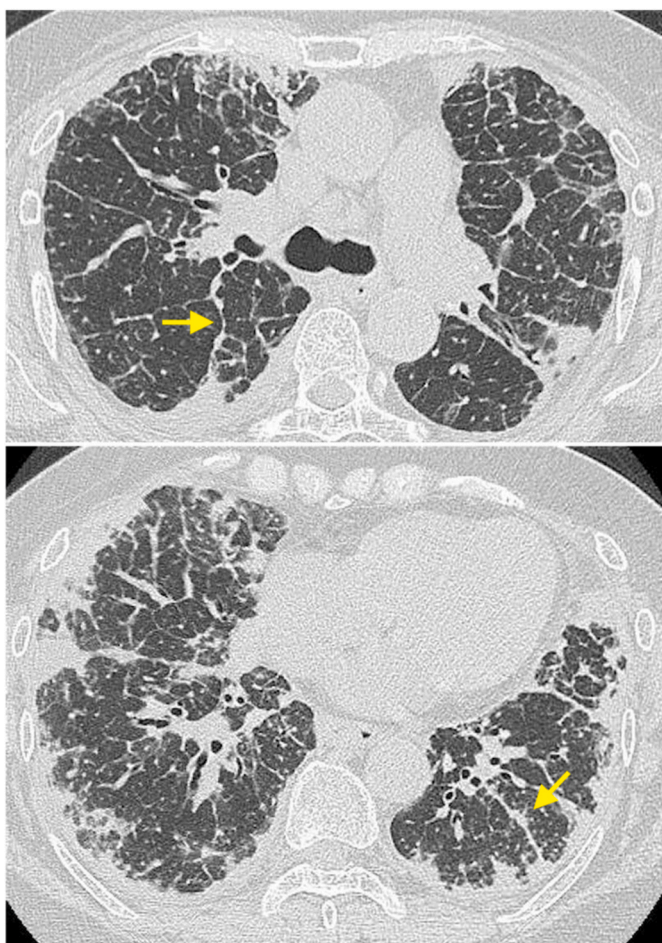


Fig. 2. Computed tomography shows diffuse nodular opacities with interlobular septal thickening (arrows) and bilateral pleural effusions.

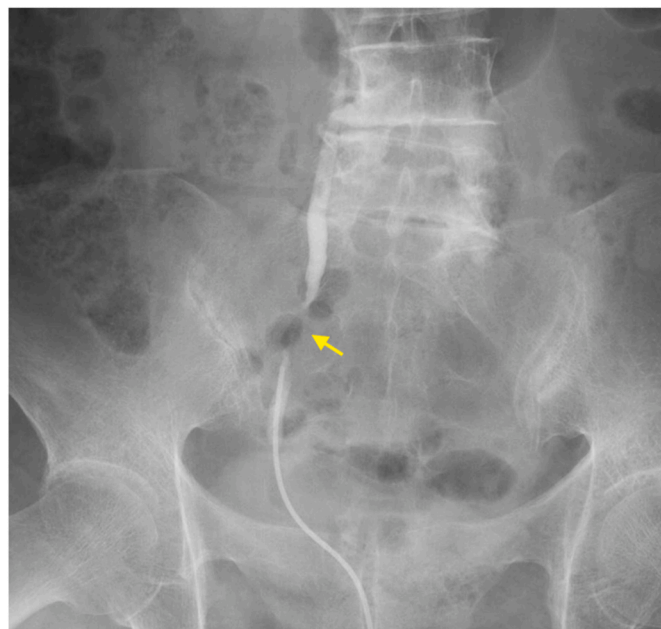


Fig. 3. Retrograde urography shows a narrowing of the right ureter (arrow).

specimens via TBLB were positive for CK-7, CK-20, and GATA-3, while negative for Napsin-A and TTF-1 (Fig. 4); thus, ruling out a primary lung adenocarcinoma. These immunohistochemistry results suggested lung metastasis of ureteral cancer. Positron emission tomography scan revealed no other origin of PLC. Therefore, we diagnosed PLC secondary to ureteral cancer, and referred her to another hospital for the specific treatment of ureteral cancer.

3. Discussion and conclusions

We diagnosed PLC secondary to ureteral cancer, and TBLB was useful for the diagnosis because PLC can imitate a respiratory failure due to other causes such as congestive heart failure, pulmonary embolism, and interstitial lung disease [1].

According to the systematic review by Klimek, which included 139 patients with PLC in 108 studies from 1970 to 2018, as the present case, dyspnea and cough are frequently observed in PLC, 59.0% and 41.0% respectively [1]. The most common origins of PLC are breast (17.3%), lung (10.8%), and gastric cancers (10.8%). In the urinary system, renal

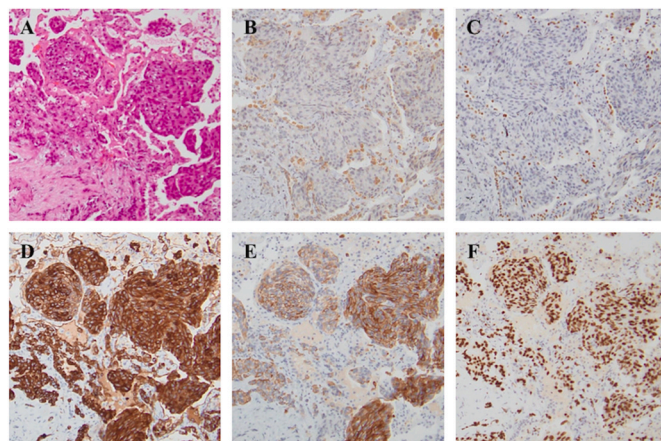


Fig. 4. Immunohistochemistry of lung specimens via transbronchial lung biopsy: (A) Hematoxylin and eosin stain, (B) Napsin-A, (C) TTF-1, (D) CK-7, (E) CK-20, (F) GATA-3.

(7.9%), prostate (5.8%), and bladder cancers (2.2%) were reported as the primary site of PLC [1]; whereas, ureteral cancer is extremely rare as the primary disease site for PLC [2]. Although occult urinary blood can be a valuable clue for the diagnosis of ureteral cancer, there was no gross or occult hematuria or urinary symptoms in the present case [3]. Thus, the CT finding of ureteral mass was the only indication of retrograde pyelography and urinary cytology.

It has been reported that BAL is highly sensitive for the diagnosis of PLC and relatively safe when compared to TBLB [4]; however, TBLB can be useful for determining the origin of PLC by immunohistochemistry. In the present case, a positive GATA-3 result was the key finding. The expression of GATA-3 is highly specific to breast or urothelial cancer [5]. Additionally, a positive CK-20 test, as is our case, suggests urothelial cancer rather than breast cancer [6]. The multidisciplinary discussion, including our pulmonologist, radiologist, pathologist, and urologist, was beneficial in reaching the diagnosis of PLC secondary to ureteral cancer.

We have reported PLC can be secondary to ureteral cancer. This case encouraged multidisciplinary discussion and a whole-body examination, including TBLB with immunohistochemistry, to determine the origin of PLC.

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Consent for publication

Written consent was obtained from the patient's husband.

Authors' contributions

HS and IH drafted the manuscript. HS, IH, KS, TO, and YK collected and interpreted the patient data. KS, RT, KN, and KK revised the manuscript.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships associated with this manuscript.

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