

F. Sema OYMAK<sup>1</sup>  
Olcay BELENLİ<sup>2</sup>  
Murat ALPER<sup>2</sup>

## Lymphangitic Carcinomatosa and Bronchorrea in a Patient With Mucoepidermoid Tumor of the Lung

Departments, of Chest Diseases<sup>1</sup> and  
Pathology<sup>2</sup>, Faculty of Medicine,  
Erciyes University , Kayseri-Turkey

Received: December 20, 1996

**Key Words:** Mucoepidermoid tumor, Lymphangitic carcinomatosa, Bronchorrea.

Mucoepidermoid tumors (METs) of the lung are uncommon tumors which are believed to derive from the excretory ducts of the submucosal bronchial glands of the proximal tracheobronchial tree. METs of the bronchial tree are histologically similar to those tumors described in the major salivary glands and usually have a favorable course (1-3). However, highly aggressive tumors may be associated with very poor prognosis (4-6). Here, an unusual case of lung MET, who presented with severe dyspnea resulting from lymphangitic carcinomatosa and bronchorrea is presented.

35 year-old male patient admitted to the hospital because of severe dyspnea. He suffered from progressively increasing weight loss, shortness of breath, non-purulent sputum expectoration and cough since last four weeks. His past medical history revealed smoking one pack/day for the last 20 years. On physical examination he was thin and mildly cyanosed with respiratory distress and breathing 38 breaths per minute. Blood pressure was 140/90 mmHg, temperature 37,2°C, pulse rate 116/min. He was anxious and had difficulty to talk. No mass could be identified in his head or neck. There were intercostal and subcostal retractions. Rhonchi and crackles were audible over both lung fields. Except appendectomy scar, abdominal examination revealed normal findings. Peripheral blood analysis results were; hemoglobin, 15,7 g/dl; white

blood cell count, 18700/mm<sup>3</sup>; platelet count, 309000/mm<sup>3</sup>. Chest radiograph obtained on admission revealed bilaterally, diffuse reticulonodular and linear shadows (figure-1). Arterial blood gas results (during 80% oxygen supply) showed both prominent hypoxemia and hypercapnea (pH: 7,44, pO<sub>2</sub>:50 mmHg, pCO<sub>2</sub>: 49 mmHg, HCO<sub>3</sub><sup>-</sup>: 31,7 meq/L, and O<sub>2</sub> saturation: 86%). Results of hepatic and renal function tests and serum electrolytes were within normal limits. Abdominal ultrasonography revealed mild hepatic enlargement. Skin test for tuberculosis (PPD) was negative. Microbiological examination of sputum was negative. Cultures failed to grow bacteria, fungi or mycobacteria. Following the admission his clinical condition continued to deteriorate and abundant increase in non-purulent sputum production (more than 200 ml/day) which had an important contribution to the development of respiratory distress was observed and a diagnostic bronchoscopy was performed. Bronchial mucosa was mildly edematous and congestive. Besides from abundant mucus secretion, innumerable, small, round (1-2 mm in diameter) and yellow-gray nodules were seen along the all visible bronchial tree, and a transbronchial biopsy was performed from left lower lobe bronchi. Following the histopathological diagnosis of mucoepidermoid carcinoma, emergent radiotherapy was planned but he died of respiratory failure at the fifth day. The autopsy suggestion was not accepted by the family.



Figure 1. The P-A chest roentgenogram shows diffuse reticulonodular and linear shadows on both lung fields.

Biopsy specimen was fixed with 10% neutral formalin and embedded in paraffin after routine tissue processing. Five  $\mu$ m sections were made.

The entire biopsy specimen was of tumoral nature by light microscopy. Large amount of mucin, forming mucin pools and filling alveolar spaces was observed. The neoplastic proliferation had a preponderance of solid or sheet-like growth. There were nests of tumor cells in mucin pools (figure-2). Stromal infiltration with inflammatory cells were also observed. Cytologic features of tumor cells were large eosinophilic cytoplasm, nuclei located centrally or peripherally and moderate atypia manifested by polymorphism, nucleomegaly, hyperchromatism and mitotic activity (one mitotic figure per 2-3 high power field). Besides from abundant extracellular mucin, intracytoplasmic mucin identified in some tumor cells by Mayer's mucicarmine stain. Tumor cells showed immunohistochemical staining with both anti-low molecular and anti-high molecular cytokeratins. There were no individual cell keratinization or squamous pearl formation. The histologic features and immunohistochemical staining pattern of the tumor cells fit well with high-grade mucoepidermoid tumor.

Histologically, METs are divided into low-grade and high-grade tumors. The majority of low-grade tumors behave in a benign fashion and high-grade classification is predictive for aggressive behavior (6). MET in

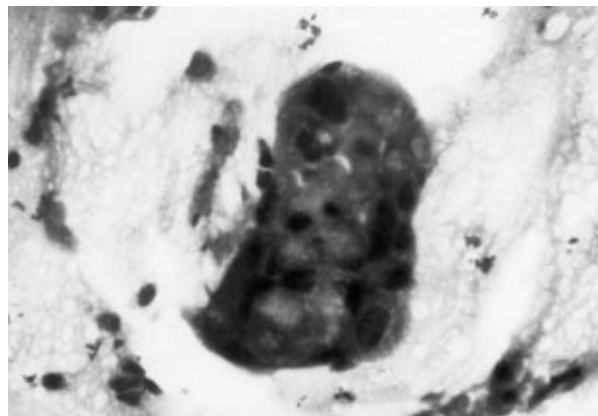


Figure 2. Light microscopic picture of the transbronchial biopsy demonstrates the nest of tumor cells in mucin pool (H&E, original magnification x 400).

this patient was a high grade one with lymphangitic dissemination.

Non-purulent bronchorrea, which has been defined as more than 100 ml/d production of mucoid sputum may be seen in patients with alveolar cell carcinoma, but it is very rare in other primary and metastatic lung cancers (7). As far as we know only one case had been reported to have lymphangitic carcinomatosa associated with bronchorrea due to metastatic colonic adenocarcinoma and, this is the first case of MET presenting with lymphangitic carcinomatosa and bronchorrea (8).

Histologically, MET is characterized by the co-existing of epidermoid, mucus-producing and intermediate tumor cells. Despite the presence of mucin-producing cells, bronchorrea has not been described in patients with lung MET. Intracytoplasmic staining of tumor cells with Mayer's mucicarmine suggests, the cause of bronchorrea in this patient was mucus production and secretion from metastasized tumor cells.

Decreases in vital capacity, diffusion capacity and pulmonary compliance are main lung function disturbances in patients with lymphangitic metastasis. In addition to these disturbances, diffuse narrowing of the airways due to submucosal tumor dissemination and abundant mucus secretion are other major factors in the development of severe dyspnea associated with hypoxemia and hypercapnia in this patient.

In conclusion, in patients who have non-purulent bronchorrea and findings suggesting lymphangitic carcinomatosa, in addition to alveolar cell carcinoma and metastatic mucus-secreting adenocarcinomas, primary lung MET should be considered in differential diagnosis.

## References

1. Healy WV, Perzin KH, Smith L. Mucoepidermoid carcinoma of salivary gland origin. *Cancer* 26:368-88,1970.
2. Dowling EA, Miller RE, Jhonson IM, Collier FCD. Mucoepidermoid tumors of the bronchi. *Surgery* 52:600-9,1962.
3. Klaesmann PG, Olson JL, Eggleston JC. Mucoepidermoid tumors of the bronchus. *Cancer* 43:1920-33,1979.
4. Metcalf JS, Maize JC, Shaw EB. Bronchial mucoepidermoid carcinoma metastatic to skin. *Cancer* 58:2556-9,1986.
5. Wolf KM, Mehta D, Claypool W. Mucoepidermoid carcinoma of the lung with intracranial metastases. *Chest* 94:435-8,1988.
6. Yousem SA, Hochholzer L. Mucoepidermoid tumors of the lung. *Cancer* 60:1346-52,1987.
7. Keal EE. Biochemistry and rheology of sputum in asthma. *Postgrad Med J* 47:171-7,1971.
8. Shimura S, Takishima T. Bronchorrea from diffuse lymphangitic metastases of colon carcinoma to the lung. *Chest* 105:308-10,1995.