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LETTERS TO THE EDITOR

# Recurrent respiratory infection and epithelial-myoepithelial carcinoma of the lung. A common presentation with a rare etiology

Dear Editor,

The tracheobronchial submucosal glands represent the pulmonary counterpart of the mucous membrane minor salivary gland system of the head and neck. These glands are rare sources of neoplasms and in these cases histologic similarity to neoplasms derived from the salivary glands becomes apparent [1]. Electron microscopic and immunohistochemical evaluation confirms the similar nature of these neoplasms [2]. Epithelial-myoepithelial carcinoma (EMC) is a rare low-grade salivary gland neoplasm with only 9 reported cases in the lung including our case [3].

A 62-year-old man was admitted with fever and recurrence of respiratory tract infection. Chest x-ray showed a shadow in the right middle lung field. A subsequent CT scan of the chest showed a right middle lobe mass with enlarged hilar lymph nodes. That lobe was completely occluded at bronchoscopy due to a smooth, tan mass that was extremely hemorrhagic, preventing a biopsy. Cytology of the bronchoalveolar lavage and sputum were negative for malignancy; also a bone scan was normal. The patient underwent right lobectomy with lymph node dissection. The cut surface of the lobectomy tissue revealed a circumscribed, solid, yellow-grey mass measuring 0.7×0.6×0.4 cm, which was attached to the bronchus of the middle lobe. Histological sections showed a uninodular, well-circumscribed, non-encapsulated neoplasm. The bronchial epithelial surface was intact covering the luminal surface of the mass. There was neither wall invasion of the bronchus nor of the pulmonary parenchyma. The adjacent adipose tissue was also intact. The neoplasm was composed of both solid and duct-like areas. Solid areas consisted of polygonal cells with pale or clear cytoplasm and uniform, ovoid nuclei that surrounded the tubular formations which had an inner layer of cuboidal or low columnar cells with eosinophilic cytoplasm and centrally located nuclei. No areas of necrosis, coagulation, vascular invasion or lymphatic vessel distension were identified. No lymph node metastasis was observed. The cells lining in the inner layer of the tubular formations were positive for cytokeratin and negative for vimentin, MSA, and S-100 protein. The cells lining in the outer layer of the tubular formations, consisting the solid areas of the neoplasm, were positive for vimentin, MSA, and S-100 protein and negative for cytokeratin. Both types of cells had no immunoreactivity for CEA, chromogranin, NSE, and CD56. The proliferation fraction as measured by Ki-67 antigen immunoreactivity was between 2 and 5% in the cell population of the neoplasm.

The postoperative course was uneventful and the patient was discharged on the 5th postoperative day. The patient is free of disease 12 months after surgery, without adjuvant chemotherapy or radiation therapy.

Little is known about the natural history of pulmonary EMCs, owing to the short-term follow-up of the reported lung EMCs cases (6-36 months). Considering the pathological similarity of the salivary and lung EMCs, the time that salivary EMCs recur or metastasize can also be expected for pulmonary EMCs cases. Salivary EMCs have a mean recurrence period of 5 years and mean metastasis period of 15 years, presenting usually as multiple pulmonary nodules [4]. These potentially malignant tumors should be removed. Lobectomy or pneumonectomy seem to be the treatment of choice, although sleeve resection can be beneficial in some cases.

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# Tissue markers of hypoglycemia: insulin growth factor-II and E-domain of pro-insulin growth factor-II expression in solitary fibrous tumor of the pleura

Dear Editor,

Solitary fibrous tumor of the pleura (SFTP) originates from submesothelial fibroblasts. It arises from the visceral, parietal or mediastinal pleura as localized, pedunculated and well circumscribed mass protruding into the pleural cavity [1]. The association of SFTP with hypoglycemia is referred to as Doege-Potter syndrome. This syndrome is described in up to 5% of patients with SFTPs [2,3]. Forty-eight patients with SFTPs were diagnosed at the Institute for Pulmonary Diseases and Tuberculosis, Clinical Centre of Serbia in Belgrade during the last 15 years. We report two of them (4.6%) with Doege-Potter syndrome. Two females (68 and 55-year-old) presented with a large pleural tumor in the right hemithorax as seen in chest X-rays, ultrasound and thoracic CT scan. They suffered from impaired consciousness with worsening dyspnoea and right-sided chest pain. Endocrine function showed an extremely reduced blood glucose level (1.1 and 0.2 mmol/L), while insulin, C-peptide, glucagon, growth hormone and catecholamines were within normal range. Percutaneous FNA of the tumor in both patients was suggestive of SFTP. The first patient declined surgery and opted for conservative treatment with i.v. glucose, while the second was operated and the tumor was resected. On the first postoperative day blood glucose returned to normal levels.

Histologically, hypercellular and hypocellular areas alternated. Small uniform sized spindle- and round-shaped tumor cells with scanty cytoplasm and without nuclear pleomorphism and mitoses were surrounded by thick to thin bundles of collagen. Prominent thin-walled blood vessels were branched, resembling hemangiopericytoma.

The tumor cells exhibited strong, uniform and diffuse cytoplasmatic vimentin, CD34 and bcl-2 positivity. According to the histological pattern and immunoprofile, both tumors were diagnosed as benign SFTPs. Dot-like immunostaining of insulin growth factor-II (IGF-II) was visualized in the vast majority of the spindle tumor cells. In the Goldgi area of the tumor cells the E-domain of pro-IGF-II was significantly more positive in the first than in the second tumor.

Non-islet cell tumor hypoglycemia (NICTH) causes reduced glucose level as paraneoplastic symptom. The majority of NICTHs are SFTPs. IGF-II and E-domain of pro-IGF-II production and secretion can be confirmed immunohistochemically as the cause of hypoglycemia [2]. Intrathoracic mass associated with NICTH should be considered as SFTP and pathologically confirmed by searching IGF-II and E-domain of pro-IGF-II on percutaneous aspiration biopsies and surgical tissue specimens. Expression of the E-domain of pro-IGF-II in tumor cells of solitary fibrous tumor originating from the pleura was not reported before as a cause of hypoglycemia according to the available literature.

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