Endobronchial primary pulmonary meningioma

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Meningiomas are common, usually benign slow-growing neoplasms of the central nervous system (CNS) comprising approximately 15% of intracranial neoplasms. Primary ectopic meningiomas are exceedingly rare extracranial or extraspinal tumors of controversial origin. Although usually limited to the head and neck region or to the paravertebral soft tissues, there are 38 reported cases of primary pulmonary meningioma (PPM) in the English literature. All were benign, except 2 that were malignant, however, none of the previously reported cases of PPM was endobronchial.[1-5] We report the first endobronchial benign PPM case who is a 34-year-old woman with a complaint of hemoptysis.

A 34-year-old woman was admitted to the Chest Diseases department with complaints of hemoptysis for approximately 15 days, and no other symptoms. Her past medical history was significant for a previous episode of hemoptysis 2 years prior, and a history of anemia. She also had a 2 pack-year history of smoking, but denied having tuberculosis or recent tuberculosis contacts. On admission, she was in good general health with normal vital signs: 110/70 mm Hg BP, 37^{0}C body temperature, 72/minutes heart rate, and 20/minutes respiratory rate. Physical examination showed diminished breath sounds at right lower zone. Routine laboratory showed a hemoglobin of 8.4 mg/dl (normal: 14-15) and a hematocrit of 28.4% (normal: 38-42). Serum iron, direct and total iron binding capacity and ferritin values were also decreased. Spirometry and arterial blood gases were normal with a forced expiratory volume in one second: 91% predicted, forced expiratory volume in one second/forced vital capacity: 0.86, and arterial oxygen saturation: 96.3%. Initial chest roentgenogram showed a sharp edged central density with reduced right lung volume. This was followed by CT imaging, which showed an endobronchial mass in the intermediary bronchus (**Figure 1a**) with atelectasis. Fiberoptic bronchoscopy showed a bright-red, smooth surfaced intrabronchial mass which was totally obstructing the intermediary bronchus (**Figure 1b**). The right upper lobe and left bronchial systems were normal. Two attempted transbronchial biopsies yielded nondiagnostic material. The patient was transferred to thoracic surgery department and a bilobectomy (right middle and lower lobes) with mediastinal lymph node dissection was performed. Gross examination of the excised lung showed grey-white, firm tumor with ill-defined borders measuring 2.5x2x2 cm in diameters. The tumor was completely obstructing the lumen of the intermediary bronchus. On cut surface, there were cystic areas containing yellowish mucoid material and an atelectatic middle lobe distal to the tumor. The tumor was unencapsulated, and well demarcated from the surrounding lung parenchyma. Histologically it contained whorls and bundles of bland polygonal to fusiform cells with oval to round nuclei and dispersed chromatin. The overall features of this tumor were benign, with minimal mitosis, nuclear atypia, and absence of necrosis (**Figures 1c & 1d**). Immunohistochemical profile demonstrated strong and diffuse positive staining with epithelial membrane antigen and vimentin, and negative staining for pancytokeratin. There was also negative staining for estrogen receptor, S-100 protein, HMB-45 (melanocytic antigen), chromogranin, synaptophysin, smooth muscle actin, desmin, CD68, CD34, glial fibrillary acidic protein while a focal positive staining for neuron specific enolase and positive staining for progesteron receptor were seen. The combination of histological and immunohistochemical features of this case lead to the diagnosis of meningioma. Postoperative cerebral and spinal magnetic resonances imaging were normal, and this case was diagnosed as PPM. The patient presented no recurrence after an 8-month follow-up. Primary ectopic meningiomas are exceedingly rare tumors. Although they are usually limited to the head and neck region or to the paravertebral soft tissues, rare cases are found to be in the lung.[1-5] Since the lung is the most common site of metastatic CNS meningioma, PPM should be considered as the definite diagnosis only after a full work up confirms the exclusion of an intracranial or intraspinal primary.[2,4] There were no clinical signs and no evidence on magnetic resonance imaging of a primary CNS meningioma in our case. Although most PPM cases had no pulmonary symptoms and were discovered as incidental intraparenchymal nodules during routine chest roentgenogram,[4,5] our case was admitted with symptom of hemoptysis probably due to the intrabronchial localization of her disease. Most cases express meningothelial, fibroblastic, or transitional growth patterns.[2] Our case expressed a meningothelial pattern. Some cases, but not ours, may contain psammoma bodies.

In conclusion, PPM is a very rare and mostly benign tumor which may mimic any pulmonary tumor especially metastasis. There is no specific clinical or radiological finding and resection is required for both diagnosis and treatment. Although most of the cases are asymptomatic and presented as an incidental intraparenchymal nodule, PPM can present with symptoms of an endobronchial tumor as seen in our case.

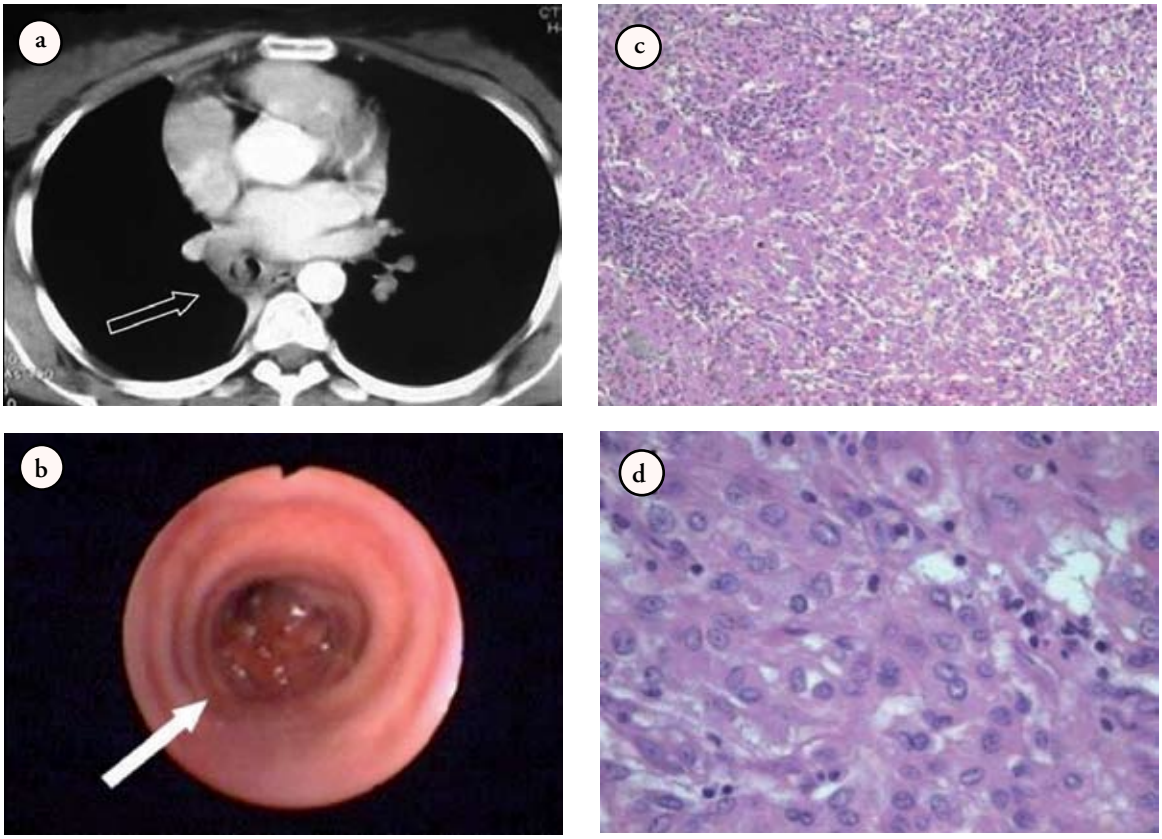

Figure 1 - **a**) CT of the thorax showing an endobronchial mass in the intermediary bronchus (arrow), **b**) Fiberoptic bronchoscopy view from the intermediary bronchus, (bold arrow). **c**) Medium magnification photomicrograph (hematoxylin and eosin) of the tumor showing epithelioid to spindle-shaped cells with ill defined cell borders arranged in whorls, **d**) High magnification photomicrograph (hematoxylin and eosin) showing syncytial aggregate of cells with bland histological features, abundant eosinophilic cytoplasm and lack of nuclear atypia, and mitotic activity.

Acknowledgement. *The authors thank to Professor Dr. Philip Cagle and Dr. Enrique M. Gomez (Department of Pathology, Baylor College of Medicine, Houston, TX, USA), Professor Dr. Buge Oz (Department of Pathology, Istanbul University Cerrahpasa School of Medicine, Istanbul, Turkey) and Dr. Sevinc Keser (Department of Pathology, Dr. Lutfi Kirdar Kartal Training and Research Hospital, Istanbul, Turkey) for their help and comments in the histopathological evaluation of the case.*

Received 24th May 2008. Accepted 15th September 2008.

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