Endobronchial Hamartoma Subtotally Occluding the Right Main Bronchus and Mimicking Bronchial Carcinoid Tumor

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Abstract: Hamartomas are very rarely identified as an endobronchial lesion. Herein, we describe a peculiar case of a 55-year-old woman with persistent cough and increasing dyspnea and radiological detection of a solid lesion subtotally occluding the main right bronchus. Despite the radiological and radiometabolic (18-fluoro-2-deoxy-d-glucose positron emission tomography/computer tomography scan) features were highly suspected for bronchial carcinoma, the definitive diagnosis after endoscopic removal was indicative of an endobronchial hamartoma. When considering differential diagnosis of an endobronchial lesion, the physicians should take firmly in mind such rare entity and, accordingly, bronchoscopy and bronchoscopic biopsy should be done as first step in management of all cases presenting with endobronchial lesions.

INTRODUCTION

Differential diagnosis of endobronchial tumors encompasses benign (e.g., hamartoma and lipoma) and malignant lesions (e.g., bronchogenic carcinoma, bronchial carcinoid (BC), metastasis, mucopidermoid, or adenocystic carcinoma), generally considering first the bronchogenic carcinoma in the differential diagnosis of malignant endobronchial lesions.

Hamartomas are rarely identified as an endobronchial lesion; in the largest review series that has been published (215 patients), only 1.4% of hamartomas had an endobronchial lesion; in the largest review series that has been published (215 patients), only 1.4% of hamartomas had an endobronchial lesion. Most of them were not associated with respiratory symptoms. Herein, we describe a peculiar case of an endobronchial hamartoma (EH) subtotally occluding the right main bronchus and mimicking BC tumor.

A 55-year-old Caucasian woman with an unremarkable past medical history presented at Emergency Department for a gradually and progressive improvement of the overall clinical conditions as well as of the radiological and endoscopic scenario was observed in the subsequent days.

After discharge, the patient underwent periodic endoscopic follow-up examinations and 12 months after operation, we observed no evidence of recurrence.

COMMENT

Pure endobronchial neoplasm (defined as the tumor mainly involving the bronchial lumen) represents a rare entity with a histological diversity. Generally, malignant diseases are more common than benign ones with non-small-cell lung cancer remaining the most predominant malignant endobronchial histology in adults followed by BC tumor (more frequently

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FIGURE 1. Radiological evaluation: chest x-ray (A) revealed a middle and lower pulmonary lobe collapse; contrast-enhanced CT-scan (B, C) showed the presence of a highly vascularized and round-shape solid lesion causing the subtotal obstruction of the main right bronchus; at $^{18}$F-FDG PET/CT scan (D) the lesion presented with a mild activity ($SUV_{max} = 2.1$), whereas no uptake was found in other areas. 

CT = computed tomography; $^{18}$F-FDG PET/CT = 18-fluoro-2-deoxy-d-glucose positron emission tomography/computed tomography.

FIGURE 2. Pathological evaluation: the polypoid tumor was constituted by a mixture of fibrous connective tissue with gelatin-cartilaginous, adipose, and fibrous components covered by bronchial respiratory epithelium (A, B).
reported in young people). Benign diseases are uncommon and, among these, EHs are a very entity with the largest series accounted for 43 EH cases.2 At careful revision of the pertinent literature, there aren’t reported specific radiological features of EH; generally, they appear as ovoid low-density endobronchial lesions,3 with a smooth edge, focal fat, or fat alternating with calcific foci (popcorn calcification).4 Such radiological findings appear substantially similar to those which may be found in BC tumors,5 this making the differential diagnosis based on radiological features only extremely challenging, as in the present case.

Treatment of the benign endobronchial tumor is almost debated; surgical approach could be considered only in very selected cases that are difficult to diagnose or remove by a bronchoscopic approach.5 On the contrary, some authors6 suggested the surgical approach as first treatment, considering that an endoscopic biopsy need to be weighed against bleeding risk especially when a BC is suspected and the lesion is located, as the present case, at the level of main bronchus.

Actually, in our opinion, the risk of bleeding in such cases could be remarkably minimized by just performing rigid bronchoscopy (instead of fiberoptic bronchoscopy). Moreover, as the present case clearly shows, a preliminary endoscopic removal may be extremely useful in all endobronchial lesions not only to solve the obstruction of the tracheobronchial tree but also to achieve a definitive diagnosis; this sometimes allows us to avoid unnecessary surgical resections for benign disease.7

In our case, the radiological features were indicative for a BC; the endobronchial lesion appeared as a round-shape lesion with slightly lobulated borders and presented punctate calcifications that may be seen in 39% of centrally located bronchial tumors.5

Moreover, the radiometabolic pattern (mild activity at 18-F FDG PET/CT scan) was also compatible with a well-differentiated BC tumor (so-called typical carcinoid), as reported in several studies.5 A DOTA-peptide PET scan could have been a very useful non-invasive diagnostic method. Indeed, considering the high accuracy in detecting BCs,6 a negative result at DOTA-peptide PET scan would be highly disproving a diagnosis of BC. Therefore, when feasible, such imaging tools should be considered in the diagnostic work-up examination of suspected BCs.

In conclusion, hamartoma may rarely occur as endobronchial tumor with a pattern of clinical symptoms and radiological/radiometabolic features that may mimic an endobronchial carcinoid tumor. Thus, a preliminary endoscopic approach may be extremely useful not only to solve the obstruction of the tracheobronchial tree but also to achieve a definitive diagnosis; this sometimes allows us to avoid unnecessary surgical resections.

REFERENCES