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Diffuse idiopathic pulmonary neuroendocrine cell hyperplasia associated with bronchiectasis: A rare case

Bronşektazi zemininde gelişen diffüz idiyopatik pulmoner nöroendokrin hücre hiperplazisi: Nadir rastlanan bir olgu

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Abstract
Diffuse idiopathic pulmonary neuroendocrine cell hyperplasia is the precursor neuroendocrine tumor proliferations comprising extended focal extraluminal nodules in whole lung occasionally or limited to the bronchial or bronchiolar walls. A 30-year old patient who underwent lower left lobectomy due to the extended saccular bronchiectasis was presented. Histopathological examination of the surgical specimen revealed diffuse idiopathic pulmonary neuroendocrine cell hyperplasia associated with bronchiectasis.
Keywords: Diffuse idiopathic pulmonary neuroendocrine cell hyperplasia, Bronchiectasis, Surgery

Öz
Nöroendokrin hücre hiperplazisi çoğunlukla havayolu mukozasında bazal membranı penetre etmeyecek şekilde ortaya çıkan nadiren tüm akciğerde yaygın odaksal ekstralüminal nodül oluşturan nöroendokrin tümör öncülü proliferasyonlardır. Bu çalışmada 30 yaşında, genişletilmiş sakküler bronşektazi nedeniyle opere edilen ve cerrahi spesmenin histopatolojik değerlendirmesinde ek olarak diffüz idiyopatik pulmoner nöroendokrin hücre hiperplazisi saptanan olgu sunuldu.
Anahtar kelimeler: Diffüz idiyopatik pulmoner nöroendokrin hücre hiperplazisi, Bronşektazi, Cerrahi

Introduction

There are numerous neuroendocrine cells within the bronchial and bronchiolar epithelium of normal lung tissue. A reactive neuroendocrine cell proliferation is observed in chronic pulmonary inflammation. Neuroendocrine cell hyperplasia is commonly seen without penetrating the basement membrane of the airway mucosal. Diffuse idiopathic pulmonary neuroendocrine cell hyperplasia (DIPNECH) are the precursor neuroendocrine tumor proliferations comprising extended focal extraluminal nodules in whole lung occasionally or limited to the bronchial or bronchiolar walls [1]. Neuroendocrine cell hyperplasia and tumorlets are frequently detected in the surrounding scar tissues and bronchiectasis and emphysematous areas, and during the microscopic examination of surgical specimens or autopsy materials.

In this report, a male diagnosed with DIPNECH associated with bronchiectasis in his histopathological examination of surgical specimen and who underwent surgery due to bronchiectasis, was presented.

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Case presentation

While a 30-year-old male with a history of smoking 15 packs of cigarette/year and obesity (Body mass index: 30 kg/m^2) and not having any additional systemic disease was assessed in a center, he was transferred to us as an extended saccular bronchiectasis in the lower left lobe of his lung was detected on computed tomography of the thorax.

He did not complain of flushing, hypertensive attack, tachycardia and diarrhea. Surgical operation was planned through the complaint of hemoptysis and extended saccular bronchiectasis. Extended bronchiectasis was detected in the lower left lobe during the surgery and lower left lobectomy was performed. Mediastinal lymph node dissection was not added as malignity was not considered.

Pathology results revealed one nodular tumor with a diameter of 3 mm on the specimen of extended saccular bronchiectasis obtained by lower left lobectomy material. DIPNECH was detected in consequence of lam revision performed at the out-center hospital. No CD56+, Chromogranin A, Synaptophysin + Ki-67 5% Mitosis were found in the pathological examination (Figure 1).

The decision of applying chemotherapy or radiotherapy were not taken during the council of thoracic oncology and only follow-up was recommended. Our case was followed-up for 17 months without any complaint.

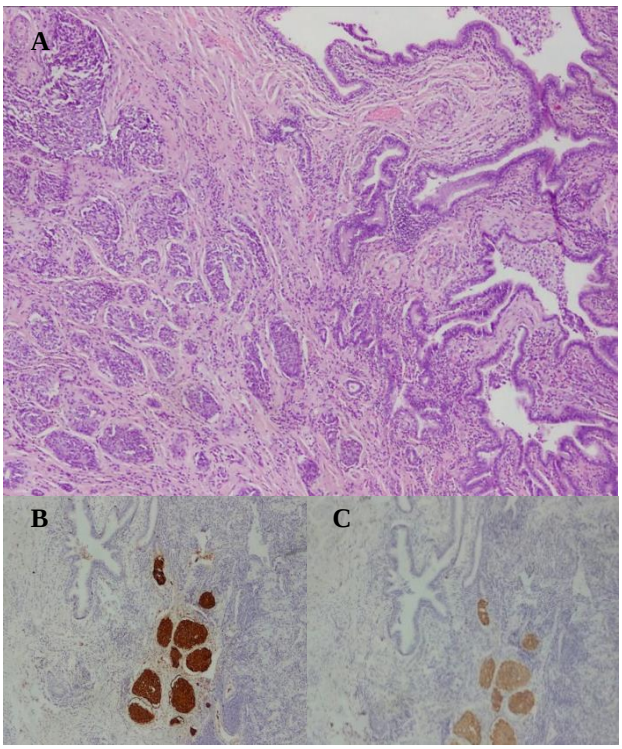

Figure 1: A: Neuroendocrine cells in small groups as well as bronchial structures. Neuroendocrine determinants are positive in the immunohistochemical study of Chromogranine (B) and Synaptophysizine (C).

Discussion

DIPNECH is a rare clinicopathological condition. Pulmonary neuroendocrine cell hyperplasia is defined as an adaptive response to chronic hypoxia in smokers or in subjects living at high altitudes [1].

Pulmonary tumorlets are defined as nodular proliferation of organoid pattern in bronchioles and originated from Kulchitsky cells. They are accidentally seen in the damaged lungs due to chronic apses, granulomatous inflammation, emphysema, infarction or similar chronic disease associated with bronchiectasis or other pulmonary sequestrations. It has been asserted that carcinoid tumorlets might be developed as a secondary response to hypoxic stress in the pathologies of bronchiectasis and sequestration [2].

It should be considered that pulmonary tumorlets must be examined as pulmonary neuroendocrine tumors and they might show intraepithelial neoplastic transformation to carcinoid. Therefore, although the primary reason of pulmonary tumorlet is benign, these patients must be followed-up for lymphatic metastasis as in patients with carcinoid [3].

Tumorlet and peripheral carcinoid are rarely associated with metastasis and lymphoid invasion. Of the typical carcinoid tumors, 4-11% tend to metastasis to the lymph nodes [4,5]. Tumorlets were classified as benign preinvasive lesions in the histopathological classification of World Health Organization (WHO) [6]. So ever, tumorlets are accepted as benign tumors, there are cases with metastasis to the lymph nodes [7,8].

The association of bronchiectasis, tumorlet and DIPNECH, which was detected in our case as well, might also trigger premalignant lesions, except the common complications, such as hemoptysis due to bronchiectasis and recurrent infections. Therefore, it should be kept in mind that early surgical therapy will prevent recurrent symptoms and further malignancies by detecting premalignant tumors at the onset of disease, in bronchiectasis patients not responding to medical therapy.

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