CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 20S (2016) 1–4

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Cushing's like syndrome in typical bronchial carcinoid a case report and review of the literature

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ARTICLE INFO

Article history:
Available online 4 February 2016

Keywords:
Cushing's syndrome
Bronchial carcinoid
Bronchoscopy

ABSTRACT

Cushing's syndrome occurred in 1–5% of cases of bronchial carcinoids. In this paper we describe a case of typical bronchial carcinoid in a nonsmoker young male with clinical manifestations mimicking a Cushing's syndrome. The patient performed chest radiograph and computed tomography. Fiberoptic bronchoscopy revealed the presence of an endobronchial mass occluding the bronchus intermedius. A rigid bronchoscopy was necessary for the conclusive diagnosis and for partial resection of the intraluminal tumor. Despite of the presence of Cushingoid features, the normal blood levels of ACTH and cortisol excluded the coexistence of a Cushing's syndrome.

1. Introduction

Bronchial carcinoids represent a small portion of all pulmonary tumors and about 1–5% of them are associated with ectopic ACTH secretion, which represents 1–10% of Cushing's syndrome cases [1–3]. Cushing's syndrome (CS) is the result of chronic exposure to increased concentration of cortisol hormone exogenous or endogenus, and it is generally associated with central obesity, metabolic syndrome, hypertension. Neuropsycological disturbances are also frequently observed, including depression, emotional irritability, sleep disturbance and cognitive deficits that can be the first manifestation perceived by family members of patients affected by CS. Treatment is based on decreasing of cortisol levels in blood, through medical or surgical approaches. Surgery is considered the main treatment for ectopic ACTH-secreting tumors. We describe a case of typical bronchial carcinoid associated with a Cushing's like syndrome.

2. Case report

A 26 year-old severe obese male with an history of anxiety over the last 5 years was admitted to our department of Respiratory Medicine referring poorly cough, fever (39 °C), sputum and shortness of breath. He was a nonsmoker and his weight, height and body mass index were 163 kg, 1.90 m and 45.15 kg × m^2, respectively. A physical exam revealed round facies, extensive purple striae on his abdomen and marked central obesity, mimicking Cushingoid features. Auscultation of the chest revealed hypophonesis and reduction of breath sound of the basal right hemithorax. Pulse was regular and blood pressure normal; the haemoglobin oxygen saturation was 91% in room air. Laboratory data showed an increase of neutrophilic count and hypertriglyceridemia. A chest radiograph showed a lift of right hemidiaphragm with ipsilateral scanty pleural effusion (Fig. 1). The clinical and radiological data suggested the diagnosis of pneumonia and the patient was treated with ciprofloxacin 400 mg iv bid and systemic steroids for 10 days. The treatment determined a substantial improvement of clinical condition, but did not modify significantly chest imagines. Subsequently the patient performed a computed tomography that revealed the presence of a solid mass in the bronchus intermedius associated with a complete atelectasis of middle and lower right lobe (Fig. 2a,b). Fibreoptic bronchoscopy revealed a pale tissue with irregular margins measuring approximately 1 cm in diameter and obliterating completely the bronchus intermedius (Fig. 3). The endobronchial biopsies showed fibrinoid material and microcalcifications in the contest of a nonspecific granulation tissue but

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http://dx.doi.org/10.1016/j.ijscr.2016.02.006

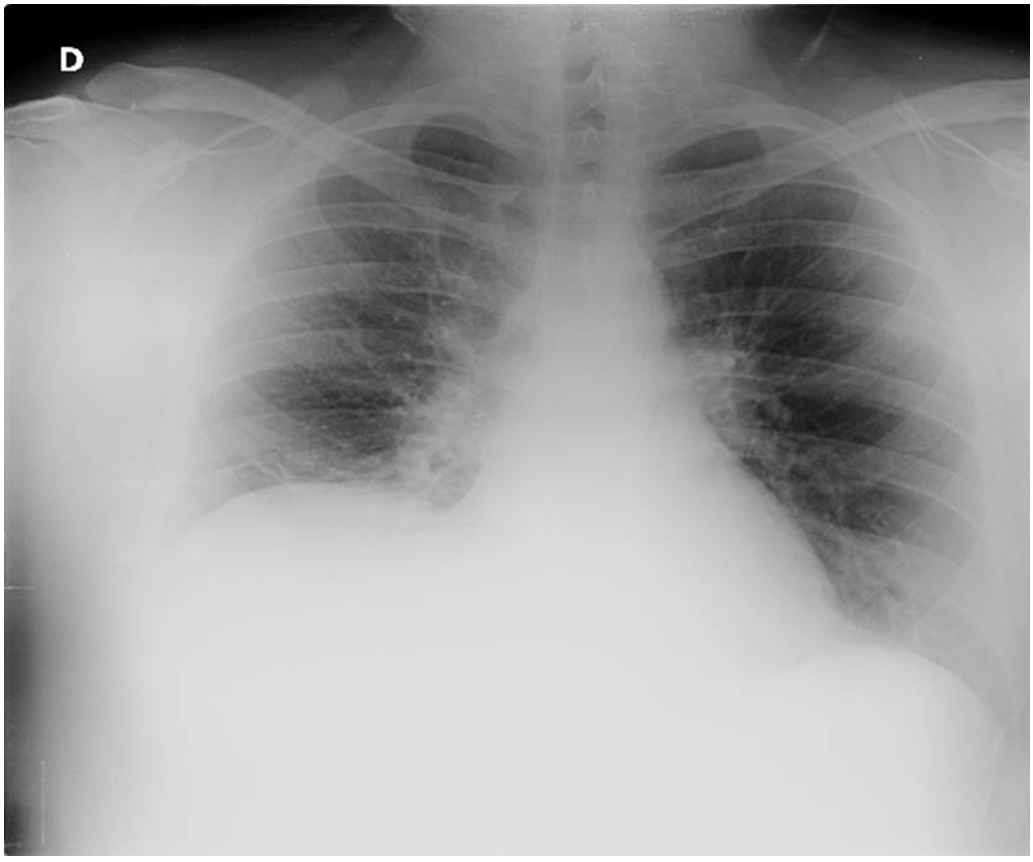

Fig. 1. Chest radiograph showing a lift of right hemidiaphragm with ipsilateral scanty pleural effusion.

were not conclusive for a diagnosis. The patient underwent a rigid bronchoscopy for diagnostic purpose and for partial resection of the intraluminal mass.

Histophatological examination of endobronchial removed samples showed a typical carcinoid tumor with a trabecular pattern and without areas of necrosis, <2 mitoses per 2 mm^2 detected by ki67 immunostaining, high CD56 and high chromogranin expression indicating neuroendocrine differentiation. On the basis of diagnosis of neuroendocrine tumor and Cushingoids features a blood test was carried out which demonstrated normal levels of ACTH and cortisol. The determination of a twenty four hour urinary free cortisol resulted normal and a high-dose dexamethasone suppression test (2 mg every 6 h for eight doses) induced a partially suppression of ACTH secretion. Moreover, the patient underwent a brain MRI that excluded the presence of a pituitary microadenoma.

A flexible bronchoscopy performed two months later showed a complete recanalization of bronchus intermedius, with a partial unblocking of middle and lower bronchi (Fig. 4). Surgical resection was considered as therapy of choice, but bilobectomy was not performed because of elevated intraoperative risk associated to the severe obesity and respiratory failure. The patient was adviced a dietary treatment and a conservative initial bronchoscopic treatment for tumor eradication.

Despite of prescription the patient was not able to lose weight (just 10 kg in one year), he still presents persistent psychiatric disorders and severe obesity. Two rigid bronchoscopies were performed during the year to avoid the local growth of the tumor and

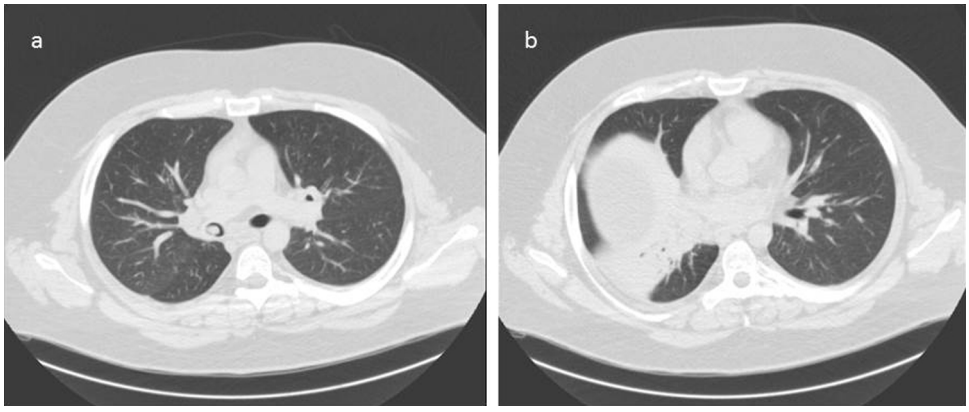

Fig. 2. Computed tomography: (a) presence of a solid mass in the bronchus intermedius; (b) atelectasis of middle and lower right lobe.