



Case Report

Persistent Hypoxemia After Bi-Basal Pneumonia in a 77-Year-Old Man: An Unusual First Presentation of Tracheobronchopathia Osteochondroplastica

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Abstract

Tracheobronchopathia osteochondroplastica is a rare but benign disease affecting the tracheobronchial tree. The main symptoms are productive cough, wheezing/stridor-dyspnea, and hemoptysis. We report the case of a 77-year-old man, who was admitted to our intensive care unit (ICU) for hypoxemia secondary to bi-basal-pneumonia, treated by high-flow nasal cannula alternated with non-invasive ventilation. Swabs, urine antigens, and sputum cultures were negative for viruses, bacteria, mycobacteria, and fungus. Despite antibiotics for 7 days, severe hypoxemia persisted, so a bronchoscopy was performed leading to the unexpected diagnosis of tracheobronchopathia osteochondroplastica. The evolution was favorable under respiratory physiotherapy, bronchial aspirations by fiberoptic bronchoscopy, and non-invasive ventilation. The patient was discharged from ICU after 13 days and from hospital 6 days later, thereafter the course was uneventful.

Keywords: Persistent Hypoxemia; Tracheobronchopathia Osteochondroplastica; Bi-Basal-Pneumonia

Introduction

A 77-year-old man, with seven days complain of bronchitis, was admitted to the Emergency Room (ER) for productive cough with yellowish sputum complicated by dyspnea and hypoxemia. His medical history was unremarkable apart arterial hypertension treated by amlodipine and losartan and hypercholesterolemia treated by red yeast rice. He also reported a mild smoking history with 5 pack-year. No chronic lung disease was reported until this hospitalization.

At the time of ER admission, he did not report fever, shivering or hemoptysis. The vital signs were normal (heart rate 83/min, blood pressure 130/70mmHg) but the patient was sub-febrile

(37.7°C), oxygen saturation was 82% on room air and therefore 5L/min oxygen was administered. Some left basal crackles were heard at the pulmonary auscultation. Chest X-Ray showed discrete bi-basal infiltrates. Laboratory results exhibited a marked CRP elevation (315 mg/L), a mild hyperleukocytosis (9.580 G/L but 8.191 G/L neutrophils), and hypoxemia on blood gases analysis (pH 7.45; PaCO₂ 35 mmHg, PaO₂ 60 mmHg). The patient was transferred to the Pneumology ward and empirically treated with Amoxicillin-clavulanic acid after obtaining sputum and blood culture samples.

Three days after admission, the patient developed acute respiratory failure with marked hypoxemia, requiring admission to the intensive care unit (ICU) and initiation of high-flow nasal cannula (HFNC) oxygenation. In this context, clarithromycin was added to the initial antibiotic to cover a possible atypical germ.

HFNC therapy was continued and alternated with non-invasive ventilation, but invasive mechanical ventilation was not necessary. Respiratory physiotherapy was performed twice a day, the patient was mobilized (active range of motion exercises including transfers from bed to the chair). While the clinical condition and the radiological images improved, the productive cough persisted as well as high oxygen demand.

On ICU admission, the patient was oriented, calm and alert. Glasgow Coma Scale score was 15/15.

His temperature was measured at 38,1°C.

Blood pressure was 156/73mmHg without vasopressor support; heart rate was 86/min. There was no mottling and skin recoloration time was less than 3 seconds. Saturation was measured at 98% under HFNC at 50L/Min 100% FiO₂. His respiratory rate was elevated at 26/minute.

He had a productive cough and bi-basal crackles.

The abdomen was soft, nontender and painless; and no mass was palpable.

Just before admission to ICU, and under 10L/min oxygen, blood gazes showed: pH 7.46, paO₂ 54 mmHg, pCO₂ 37 mmHg, and normal lactate level (1.1 mmol/L).

Laboratory results, including renal and liver functions, were normal except a significant inflammatory syndrome (CRP 189 mg/L) and white blood cells 9.700 G/L (maximal normal range 9.5 G/L).

Chest X-Ray confirmed pneumonia of both lower lobes (figure 1).



Figure 1: Chest X-Ray showing bi-basal lung infiltrates.

Nasopharyngeal swabs were negative for SARS-CoV-2, chlamydia pneumoniae and mycoplasma pneumoniae. Urinary legionella and pneumococcal antigens were also negative.

Endotracheal cultures were negative for bacteria, mycobacteria and fungus. Blood cultures on arrival in the emergency room and during ICU stay remained negative.

Amoxicillin-clavulanic acid and clarithromycin were discontinued after 7 days, given the regression of the biological inflammatory syndrome.

Given persistence of a productive cough and HFNC requirements, a fiberoptic bronchoscopy was performed, showing multiple nodules on the anterior wall of the distal trachea and proximal mainstem bronchi with a lot of purulent secretions (figure 2, a-b), leading to the diagnosis of tracheobronchopathia osteochondroplastica (TBO).

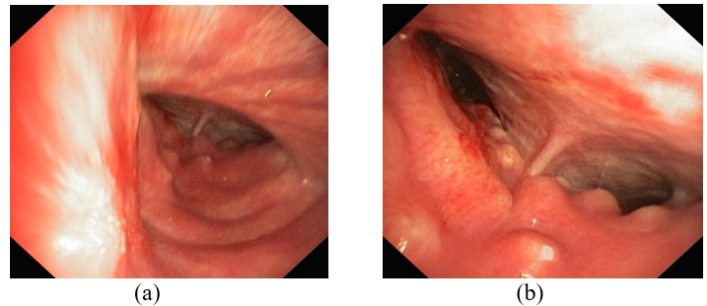


Figure 2: Fiberoptic bronchoscopy showing multiple nodules on the anterior wall of the distal trachea and proximal mainstem bronchi.

Pulmonary embolism was excluded by injected thoracic CT but also demonstrated some tracheal nodules (figure 3).

Transthoracic echocardiography showed preserved left systolic function with no evidence of pulmonary hypertension. Bubble study excluded a patent foramen ovale.

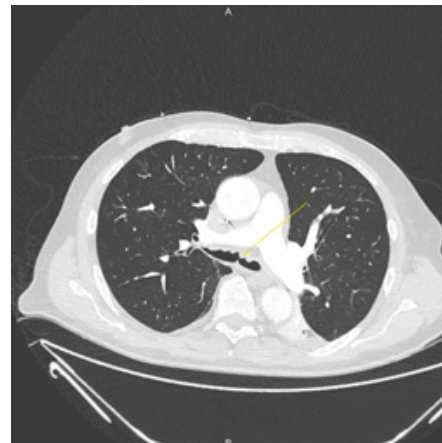


Figure 3: High resolution pulmonary CT-scan showing nodules (yellow arrow).

A search for an extra-thoracic shunt using technetium scintigraphy was also negative.

The evolution of the patient was favorable after multiple sessions of respiratory physiotherapy, bronchial aspirations by fiberoptic bronchoscopy and non-invasive ventilation.

The advantage of bronchoscopy lays in bronchial toilette, which enables trapped secretions to be aspirated.

Oxygen requirements gradually decreased, enabling the patient to be weaned from HFNC and to be transferred to the pneumology ward after 13 days.

The patient was discharged from the hospital 6 days later.

Four months later, a follow up pneumology consultation noted the disappearance of all previous respiratory symptoms.

Discussion

Tracheobronchopathia osteochondroplastica is a benign but probably underreported condition affecting the tracheobronchial tree in adults [1]. First described by Wilks in 1857, it is characterized by the development of osseous and cartilaginous nodules within the submucosa of the anterior part of the trachea and bronchi, typically sparing the posterior wall.

The multiplication of nodules progressively alters the anatomy of the tracheobronchial tree, impairing adequate clearance of secretions and leading to obstructions and respiratory infections.

To date, the etiology of TBO remains largely unknown, and although several authors reported associations with multiple comorbidities, no conclusive evidence could be made for TBO. In addition, no clear relationship could be established between smoking and the appearance of TBO [2]. The diagnosis is usually established between the fourth and seventh decade, with a mild male predominance (ratio 1.73) [1-3].

While patients can be asymptomatic (13%) [1,2], being diagnosed during intubation for an elective surgery or incidentally during diagnostic procedures for unrelated diseases, the most frequently reported symptoms are a chronic productive cough (53%), wheezing/stridor-dyspnea (35%), hemoptysis (23%), recurrent lower respiratory tract infections (10%), intermittent chest tightness/pain (7%), and voice change (7%) [2].

Actually, TBO is not associated with a specific clinical course, and the aspecific presentation may lead to other diagnosis such as asthma [2].

The reduction in bronchial lumen associated with the presence of sticky secretions, especially during episodes of airway infection, can lead to ventilatory disturbances and even respiratory failure [4].

Chest radiography is usually normal, although atelectasis or consolidations can be observed, as well as some tracheal or mainstem bronchi abnormalities (nodular irregularities, thickening, calcification, and luminal narrowing). As expected, thoracic Ct-scan is more sensitive to demonstrate tracheal bronchial tree lesions characterized by irregular calcific or cartilaginous densities along the trachea and bronchi typically sparing the posterior membranous wall [2], but may be normal. The CT-scan may also underline the presence of stenosis/narrowing of the respiratory tract lumen.

Fiberoptic bronchoscopy is pathognomonic and is considered as the gold standard for diagnosis [1]. The typical appearance includes multiple, sessile, whitish or pale, and polypoidal or nodular lesions involving partly or totally the anterior and lateral wall of the respiratory tract but sparing the posterior membrane [2].

Pulmonary function tests, when available, are non-specific and vary drastically from patient to patient without any clear pattern [2]; ranging from an obstructive pattern of variable severity (as in our patient who exhibits a mild obstruction: FEV1 85% and FEV1/FVC 75%) to a restrictive one, passing through normal [2].

The treatment is mostly supportive, consisting of treating respiratory infections and improving clearance of secretions. Respiratory physiotherapy may help in this context. In severe cases, laser removal of all the bony and cartilaginous nodules can be performed [2,3], as well as debulking with the help of rigid bronchoscope or by surgery. To date, no treatment or technique can prevent the development or the progression of the disease.

In about ¾ of the reported TBO cases, the evolution had an indolent and non-progressive course, and intervention was finally required in patient with worsening symptomology and narrowed airway. Mortality was rarely directly attributed to TBO.

Conclusions

Although prolonged hypoxemia could be encountered in ICU patients, this rarely leads to diagnosis of TO. This also emphasizes the importance of performing a fiberoptic bronchoscopy in any etiological assessment of persistent hypoxemia because it is the easiest and, above all, the most cost-effective way to diagnose this rare entity that is TO.

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