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Mounier-Kuhn Syndrome Revealed by Respiratory Exacerbation with Associated Interstitial Lung Disease: A Case Report

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Abstract **Case Report**

Tracheobronchomegaly (TBM) or Mounier-Kuhn syndrome, is characterized by marked dilation of the trachea and proximal bronchi associated with recurrent respiratory infections. Clinical signs are not very specific. Diagnosis is mainly based on chest radiography and computed tomography, relying on measurement of the diameter or surface area of the trachea and main bronchi. Treatment is essentially symptomatic, including respiratory physiotherapy and antibiotic therapy. Surgical interventions may sometimes be required. We report the case of a 67-year-old chronic smoker in whom this syndrome was diagnosed in our Pulmonology Department at Mohammed VI University Hospital in Marrakech.

Keywords: Trachea; Mounier-Kuhn syndrome; Tracheobronchomegaly, Bronchiectasis, Recurrent respiratory infections.

INTRODUCTION

Tracheobronchomegaly, or Mounier-Kuhn syndrome, is a rare disease of debated congenital or acquired origin, characterized by significant dilation of the trachea and proximal bronchi. It is a rare condition secondary to defective development of connective tissue and smooth muscle of the trachea and bronchi, leading to tracheobronchial dilation. Clinical signs are varied and non-specific. Imaging allows a positive diagnosis and assessment of pulmonary parenchymal involvement. We report a case of tracheobronchomegaly revealed in the context of a respiratory exacerbation associated with diffuse interstitial lung disease.

PATIENT AND OBSERVATION

Patient Information:

A 67-year-old male patient with a history of chronic smoking (40 pack-years) and recurrent respiratory infections. He had never been treated for pulmonary tuberculosis.

He presented with chronic exertional dyspnea associated with morning bronchorrhea. The course was marked by progressive worsening of dyspnea, reaching stage IV according to Sadoul classification over the past four months, associated with moderate atypical chest pain, without extra-thoracic signs. The condition evolved in an afebrile context with deterioration of general status (anorexia, asthenia, unquantified weight loss).

Clinical and Paraclinical results:

Clinical examination revealed poor general condition with WHO performance status 4, afebrile (37°C), polypnea (28 cycles/min), blood pressure 110/70 mmHg, tachycardia (105 bpm), and oxygen saturation 82% on room air, improving to 90% with 4 liters of oxygen. Pulmonary auscultation revealed bilateral rhonchi.

The frontal chest X-ray showed diffuse bronchial syndrome, loss of left lung volume, mediastinal widening with deviation of the trachea to the right (Figure 1). Chest CT scan demonstrated dilation of the trachea and proximal bronchi associated with parietal tracheal diverticula. Fibrosing diffuse interstitial pneumonia was observed, associated with minimal pneumomediastinum, minimal bilateral pleural effusion, and minimal right apical pneumothorax (Figure 2).

Salivary gland biopsy showed subacute and chronic non-specific sialadenitis grade I according to

Chisholm and Mason, with no signs of malignancy. Immunological tests were normal. Bronchoscopy was not performed due to the patient's respiratory status and WHO performance status 4. Plethysmography and the 6-minute walk test could not be performed. Arterial blood gases showed severe type I respiratory failure. Laboratory tests revealed inflammatory syndrome with elevated C-reactive protein and neutrophilic leukocytosis. Sputum examination for tuberculosis bacillus was negative. Angiotensin-converting enzyme levels, phosphocalcium balance, and urinary tests were normal.

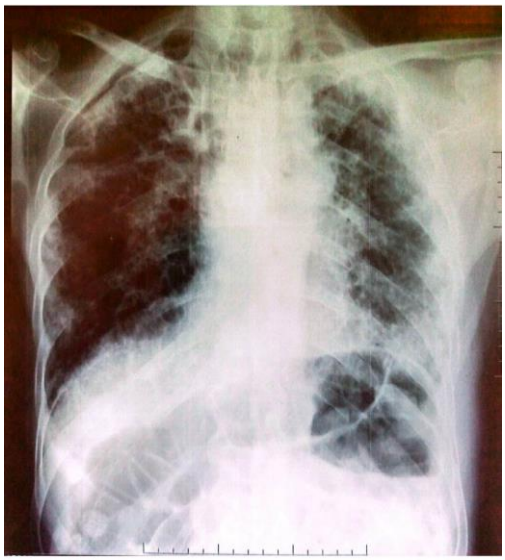

Figure 1: Diffuse bronchial syndrome, loss of left lung volume, mediastinal widening with deviation of the trachea to the right, right apical detachment; interstitial syndrome predominating on the left

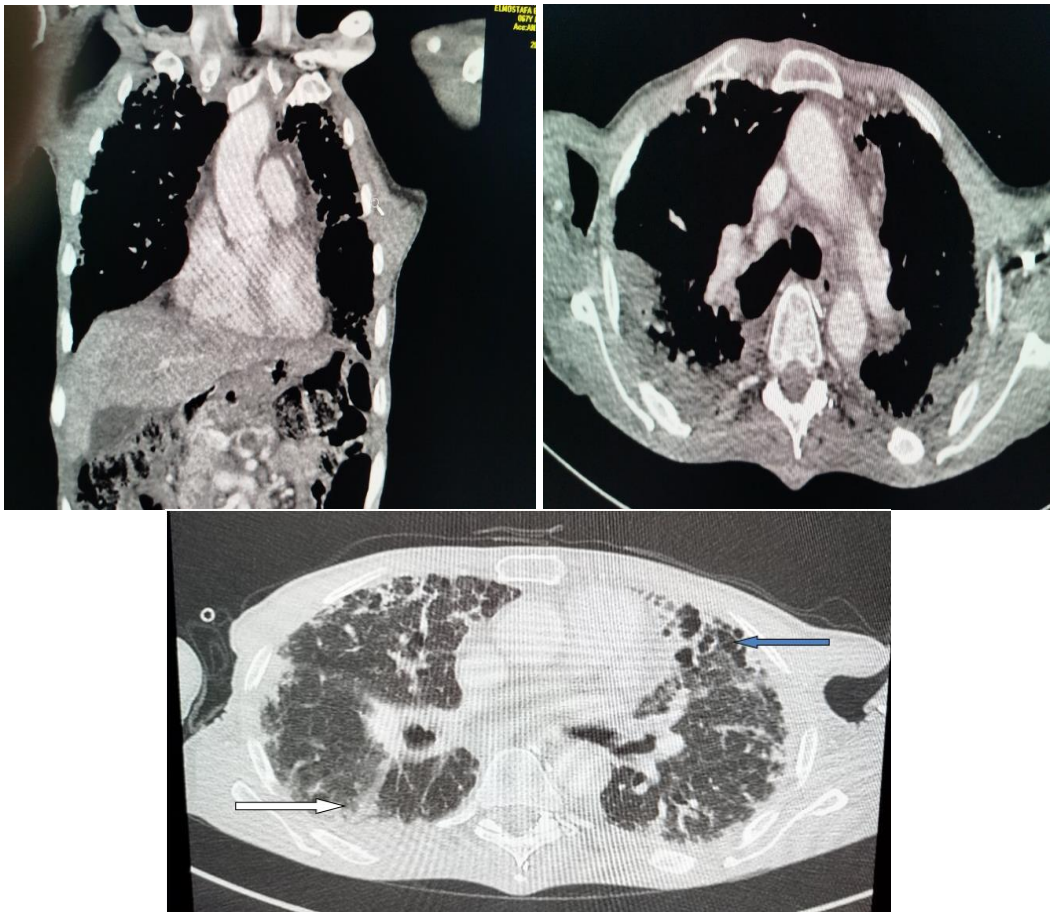

Figure 2: Chest CT scan showing tracheobronchomegaly associated with parietal tracheal diverticula; fibrosing diffuse interstitial pneumonia; minimal pneumomediastinum; minimal bilateral pleural effusion; minimal right apical pneumothorax

DISCUSSION

Mounier-Kuhn syndrome (tracheobronchomegaly, TBM) is a well-defined condition both clinically and, more importantly, radiologically. It is characterized by marked dilation of the trachea and main bronchi and is frequently associated with bronchiectasis, tracheal diverticula, and recurrent bronchopulmonary infections [1,2]. The syndrome may also be associated with nasosinusal polyposis and polymalformative features, including bilateral ptosis, epicanthus, micrognathia, and excess upper lip skin [1,3].

The first endoscopic and radiologic description was reported by Mounier-Kuhn in 1932 [4]. The term "tracheobronchomegaly" was later introduced by Katz et al. in 1962 [5]. This rare condition predominantly affects young adult males between the third and fourth decades of life [1,6], although cases have been reported across a wide age range, from infancy to elderly patients [7].

The etiopathogenesis of TBM remains uncertain. A congenital origin with autosomal recessive inheritance has been suggested, particularly due to its association with connective tissue disorders such as Ehlers-Danlos syndrome and cutis laxa in children [1,8]. However, the frequent occurrence of TBM in adults with sporadic presentation supports a possible acquired origin [1,8]. Several contributing factors have been proposed, including barotrauma related to neonatal ventilation and oxygen therapy, as well as chronic exposure to airway irritants such as tobacco smoke and environmental pollution. The underlying mechanism involves structural abnormalities of elastic and smooth muscle components of the tracheobronchial wall [9,10].

Clinically, patients typically present with chronic cough and recurrent respiratory infections. The cough is usually productive and may be associated with hemoptysis. As the disease progresses, cough effectiveness decreases due to progressive airway dilation, leading to secretion stasis and repeated infections. The disease often evolves toward chronic bronchitis, with initially preserved pulmonary function that progressively deteriorates, eventually resulting in respiratory failure [1,11].

The diagnosis of TBM is based on standardized measurements of the trachea and main bronchi. Although these measurements can be obtained on chest radiography, computed tomography provides greater accuracy [1,12,13]. In men, TBM is defined by a transverse and sagittal tracheal diameter exceeding 30 mm, and/or a right and left main bronchial diameter exceeding 18 mm and 21 mm, respectively. Corresponding threshold values in women are lower [1,14,15].

Computed tomography also plays a crucial role in assessing associated bronchial and parenchymal abnormalities [1,9]. Bronchiectasis is commonly observed, and various parenchymal lesions may coexist, including atelectasis, infections, interstitial fibrosis, or emphysema, as illustrated in our case.

The differential diagnosis includes Williams-Campbell syndrome, which is characterized by congenital cystic bronchiectasis due to cartilage deficiency in the fourth- to sixth-order bronchi. However, in this condition, the trachea and main bronchi are of normal caliber [1,5].

Bronchoscopy has limited diagnostic value due to the marked airway dilation, and diagnosis mainly relies on imaging studies. Tracheobronchography is rarely performed in current practice [12].

Management is primarily supportive and focuses on airway clearance through respiratory physiotherapy and antibiotic therapy during infectious exacerbations. In severe cases, continuous positive airway pressure or endobronchial stenting may be considered [1,16,17]. Inhaled bronchodilators and corticosteroids are generally ineffective. Surgical resection has limited utility, as the disease rarely remains confined to the central airways.

CONCLUSION

Mounier-Kuhn syndrome or tracheobronchomegaly is a very rare condition of debated congenital or acquired origin. Clinical signs are non-specific and radiological diagnosis is often straightforward. CT evaluation of central airways and pulmonary parenchyma is essential. Treatment remains mainly supportive despite attempts at surgical or interventional approaches.

Conflict of Interest: The authors declare no conflicts of interest related to this article.

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