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# OSTEOCHONDROPLASTIC TRACHEOBRONCHOPATHY: A CASE REPORT

***Ludmila Naves Marinho***
Instituto Hospital de Base do Distrito Federal - Unidade de Endoscopia Respiratória
Brasília - Distrito Federal

***Laura Pelizaro***
Instituto Hospital de Base do Distrito Federal - Unidade de Endoscopia Respiratória
Brasília - Distrito Federal

**Abstract:** Osteochondroplastic tracheobronchopathy is a rare and benign condition characterized by sessile submucosal nodules in the larynx, trachea and bronchi, predominantly on the anterolateral wall of the trachea. The pathophysiology remains unknown, however studies suggest an interaction between genetic and environmental factors, with the presence of BMP-2 identified in immunohistochemical analyses. The symptoms are varied, with 50% of cases being asymptomatic. Diagnosis is confirmed by bronchoscopy, and the disease can be classified into stages A, B and C. Complications include atelectasis and tracheal stenosis. There is no specific treatment, with therapeutic options in cases of reduced tracheal caliber, such as the use of laser or silicone molds. Approximately 55% of cases do not show progression, indicating heterogeneity in the evolution of the disease.
**Keywords:** tracheopathies, tracheal stenosis, dyspnea

## INTRODUÇÃO

Osteochondroplastic tracheobronchopathy, a rare and benign condition characterized by the presence of sessile submucosal nodules composed of cartilaginous and/or bone tissue. This disease manifests itself predominantly in the larynx, trachea and bronchi. The pathophysiology is unknown. This article seeks to report a case of the disease as it is a rare condition and discuss its clinical, pathophysiological, diagnostic and therapeutic characteristics.

## CASE REPORT

A 61-year-old male patient referred to the Respiratory Endoscopy department for investigation of chronic cough and dyspnea. Upon admission, he reported that for approximately 3 years he had progressively worsening dyspnea, sporadic dry cough and "wheezing". He denied fever, hemoptysis, weight loss, or other respiratory symptoms. He had comorbid spinal canal stenosis and knee osteoarthritis with difficulty walking. He denied smoking or using illicit substances.

According to him, he had sought medical care in a basic health unit with flu-like symptoms and a chest CT scan was performed which showed changes, and he was referred to the Respiratory Endoscopy unit for bronchoscopy.

On physical examination, he was in good general condition, lucid and oriented in time and space, hemodynamically stable. From a respiratory point of view, he was eupneic, SpO2: 95% on room air. On respiratory auscultation, he presented bilaterally discrete wheezing and on cervical auscultation, inspiratory and expiratory stridor.

An endoscopic examination was performed and the presence of multiple whitish, irregular, hardened (calcified) lesions in the trachea were observed, located throughout the trachea, on the lateral and anterior walls that extended to the right and left main bronchi.

After completion, the patient was referred to the Thoracic Surgery unit for evaluation.

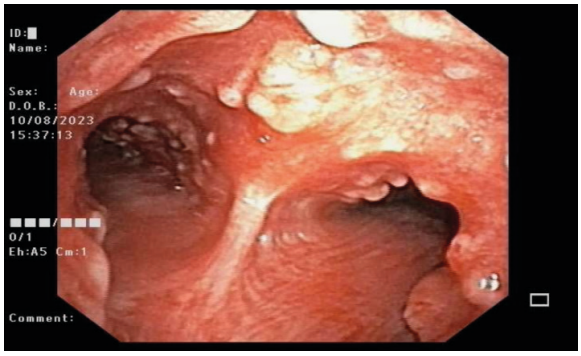


Figure 1: Bronchoscopy examination demonstrating trachea, main carina and right and left main bronchi affected by whitish submucosal nodules

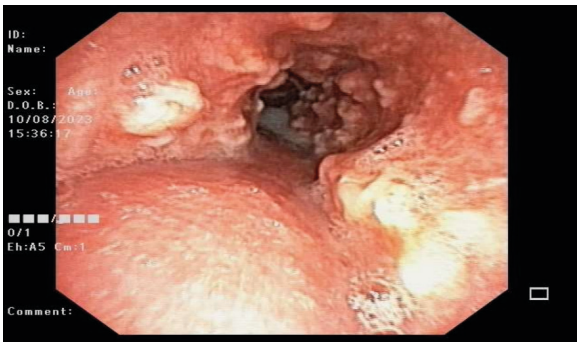


Figure 2: Bronchoscopy examination demonstrating trachea affected by whitish submucosal nodules

## DISCUSSION

Tracheobronchial osteochondroplasty is a rare and benign condition that is characterized by the presence of sessile, whitish submucosal nodules, composed of cartilaginous and/or bone tissue, giving them a hardened consistency. These nodules are distributed in the larynx, trachea and bronchi, being predominantly located on the anterolateral wall of the trachea, with a tendency to spare the membranous wall. There are few reports of laryngeal and sublotic involvement. Its anatomical distribution comprises approximately 42% in the trachea, 6% in the bronchi and 52% in both regions.

The pathophysiology of this condition remains unknown to date, although studies suggest a possible interaction between genetic and environmental factors. In an immunohistochemical analysis performed by Tajima et al. In 1997, the presence of bone morphogenic protein (BMP-2) was identified in lesions, this protein being responsible for bone formation and repair. The usual absence of this protein in the tracheobronchial tree suggests that its abnormal production may trigger the formation of the aforementioned nodules. At high concentrations, BMP-2 stimulates the expression of transforming growth factor messenger ribonucleic acid (mRNA) (TGF-β1), promoting the production of extracellular matrix proteins for chondrocytes. Leske et al., in 2001, when performing biopsies on 40 patients, observed changes such as ossification (58%), squamous metaplasia of the respiratory epithelium (48%), cartilage (38%), calcification (20%) and amyloidosis (13% ).

The symptomatology of the disease is diverse and, in approximately 50% of cases, it can be asymptomatic, and is often diagnosed incidentally during chest CT scans performed for other reasons. When symptomatic, it generally manifests itself as a productive cough due to difficulty in eliminating secretions, wheezing, hemoptysis and recurrent infections. Dyspnea is progressive, correlated with the degree of impairment and reduction in tracheal caliber.

Imaging tests, such as chest x-rays and computed tomography scans, can raise suspicions of the disease, showing irregularities and narrowing of the trachea. Bronchoscopy, however, is essential for the definitive diagnosis, allowing a more detailed assessment of the extent and severity of the pathology. In 2004, Dutau et al proposed a disease severity classification based on the extent of endoscopic lesions, divided into 3 stages:

- Stage A: Scattered nodules (large area of normal mucosa between them)
- Stage B: Diffuse nodules (no area of normal mucosa)
- Stage C: Confluent lesions (fusion of adjacent lesions)

When considering the differential diagnosis, other conditions such as papillomatosis, sarcoidosis, chondrosarcoma, hamartomas, amyloidosis, tuberculoid calcifications, dermatomyositis, scleroderma, Wegener's disease, sarcoidosis and calcified paratracheal lymph nodes must be considered, with radiological and endoscopic characteristics being crucial for differentiation.

Frequent complications include atelectasis, recurrent pneumonia and tracheal stenosis. There is no specific treatment for tracheobronchial osteochondroplasty, and in approximately 55% of cases there is no progression of the disease, as identified by Lazor et al. in a series study carried out in 2001. In case of reduction in tracheal caliber, therapeutic options such as the use of laser or the placement of silicone molds can be considered. Tracheostomy may occasionally be necessary.

## CONCLUSION

Tracheobronchial osteochondroplasty is a condition whose origin remains unknown, characterized by anomalous ossification of the cartilages of the larynx, trachea and bronchi. Symptoms arise directly from the location and degree of airway obstruction. There is no specific treatment for the disease, with clinical control being the standard approach in asymptomatic cases or with mild symptoms. Surgical removal is reserved for situations of moderate to severe obstruction. Long-term clinical follow-up is essential in these patients.

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