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Caso Clínico *Case Report*

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Bronquiolite constritiva ocupacional (?) em doente com exame físico, radiológico e funcional normal

Occupational (?) constrictive bronchiolitis with normal physical, functional and image findings

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Resumo

A bronquiolite constritiva é caracterizada por alterações das paredes dos bronquíolos membranosos e respiratórios. Estas alterações incluem um espectro de alterações que podem variar, desde a inflamação à fibrose concêntrica progressiva, com obstrução completa do lúmen bronquiolar. O diagnóstico pode ser sugerido pela história clínica e por alterações radiológicas e funcionais. No entanto, o exame físico e os exames complementares de diagnóstico podem ser normais, o que dificulta o diagnóstico, sendo necessário um elevado índice de suspeita para se sujeitar o doente a exames invasivos, tal como a biópsia pulmonar cirúrgica. Os autores apresentam um caso clínico de uma doente com quadro arrastado de tosse e dispneia, com exame físico, funcional e imagiológico normais, cujo estudo exaustivo veio a revelar o diagnóstico de bronquiolite constritiva.

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Palavras-chave: Bronquiolite constritiva, óxido de ferro.

Abstract

Constrictive bronchiolitis is characterized by alterations in the walls of membranous and respiratory bronchioles. These changes lead to concentric narrowing or complete obliteration of the airway lumen. Suspicion of possible bronchiolar disorders may arise from clinical, functional, and radiologic findings. However, constrictive bronchiolitis may be present even with normal physical, functional and image findings, which turns the diagnosis difficult. A high index of suspicion is necessary to justify invasive tests that lead to pulmonary biopsy. In this report, we describe a patient with cough and dyspnoea, with normal physical, functional and image findings, whose work-up led to the diagnosis of constrictive bronchiolitis.

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Key-words: Constrictive bronchiolitis; iron oxide.

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BRONQUIOLITE CONSTRITIVA OCUPACIONAL (?) EM DOENTE COM EXAME FÍSICO, RADIOLÓGICO E FUNCIONAL NORMAL

Sandra Figueiredo, António Morais, Adriana Magalhães, Conceição Souto Moura João Almeida, Isabel Gomes

Case report

The authors present a case report of a 26 years old Caucasian female that was referred to hospital with symptoms of cough and dyspnoea at the work place, which had been present for 2 years. She had no history of tobacco use or previous respiratory disease. She had worked with copy-machines for several years until 2 months before the first visit. Symptoms improved when she was away from the work place and, at the time of observation, she had no respiratory symptoms and presented a normal physical examination, including pulmonary and cardiac auscultation.

Laboratory studies, including blood tests, RAST and D-dimers, were within normal range. Lung function tests with spirometry, lung volumes and DLCO were normal. Arterial blood gases were also normal. A chest radiograph (Fig. 1) revealed no abnormalities. Subsequently, a High Resolution Computerized Tomography (HRCT) of the chest was performed (Fig. 2A, B and C) which was normal, as well.

The patient returned to the work place for a fortnight just to perform a PEF curve with morning and evening measurements, but it did not suggest occupational asthma. A methacoline challenge was undertaken and was negative. She managed 517 metres du-



Fig. 1 – Chest radiograph

ring the 6-minute walk test with no significant drop in saturation (3%).

Ten months after the first visit, the patient was still away from work, without respiratory symptoms outside the context of common colds, and her physical examination was still normal, as well as pulmonary function tests and methacoline challenge. So as to exclude any involvement not detected by the previous tests, fiber optic bronchoscopy was then performed, with unremarkable endoscopic findings. However, the bronchoalveolar lavage done in the right middle lobe revealed a lymphocytosis (32,4%) with a normal CD4/CD8 ratio.



Fig. 2 – (A,B and C) High-resolution computed tomography of the chest



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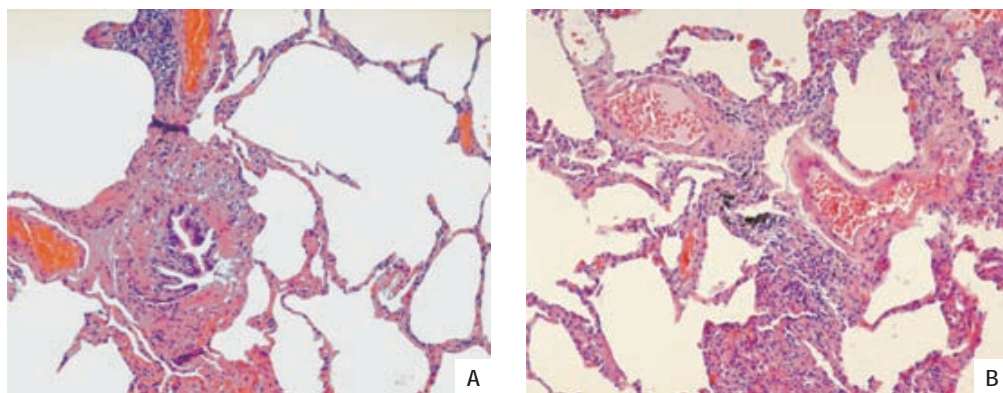


Fig. 3 – (A) Concentric fibrosis surrounding the bronchiole lumen (HE 400x); (B) The bronchiole is infiltrated by chronic inflammatory cells (HE 400x)

At this point, a lung specimen was needed and surgical lung biopsy was recommended to the patient. Histopathological assessment (Fig. 3) revealed “Lesions of chronic cellular bronchiolitis, fibrosis and smooth muscle proliferation of the bronchiolar wall, suggestive of *constrictive bronchiolitis*...”

Treatment with oral deflazacort was started and maintained for 6 weeks, but it was discontinued since the patient was clinically stable and possible benefits would not balance long-term side-effects. She has been medicated with an inhaled corticosteroid since then. The powder composition of the copy-machine was evaluated and it included iron oxide, which has been associated with bronchiolitis. No other suggestive agent was reported by the patient in her environment, including of protein origin. She has been kept away from copy-machines ever since and she has been clinically and functionally stable for a 22 months follow-up period.

Discussion

Mineral dust exposure is classically associated with restrictive lung disease. However

mineral dusts can also produce abnormalities in the small airways with airway obstruction^{1,2}. The deposition of inhaled dust around small airways appears to lead to a repair mechanism with an inflammatory response that may induce bronchiolar and alveolar epithelium injury¹⁻⁴. This process may result in complete recovery or may be characterized by proliferation of granulation tissue with concentric narrowing or complete obliteration of the airway lumen, which, in constrictive bronchiolitis, occurs at the level of membranous and respiratory bronchioles. Among those subjects exposed to mineral dusts, individual susceptibility to this bronchiolar lesion may determine by individual ability to clear these particles from the airways¹.

Iron oxide has been implicated in the etiology of constrictive bronchiolitis and it was present in the toner composition of the copy-machines, at the patient's work place. Although direct implication can not be assured, it is possible that it triggered the inflammatory response for the initiation of the lesions in this case.



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Clinical manifestations of constrictive bronchiolitis depend upon the type, extent or severity of the lung injury and tend to progress with time, with increasing dyspnoea, wheezing and cough, but sputum production can also be present. This woman was symptomatic only at the work place.

Radiographic abnormalities may be absent or include progressive increase in lung volume on serial radiographs or marked heterogeneity of lung density in HRCT scan. In this case, HRCT in expiration was not performed previously to pulmonary biopsy, and it could have shown a mosaic pattern suggestive of heterogeneity. However, to perform this exam in this patient, a high index of suspicion would also be needed and, even with a positive finding, it would not preclude invasive diagnostic procedures. The interest of perfusion and ventilation isotope lung scans has been mentioned in some forms of bronchiolitis⁵, but findings are expressive when patients have significant functional impairment. In fact, in bronchiolitis, pulmonary function tests typically

present an obstructive defect with hyperinflation that was not present in this patient. In our opinion, the remarkable feature in this case is that constrictive bronchiolitis was present when different non-invasive tests for evaluation of small airway disease were normal, which turned the diagnosis difficult, and a high index of suspicion was necessary to justify the invasive procedures that lead to lung biopsy.

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