

Bronchostenosis with atelectasis of the middle lobe of the right lung as one of rare complications of sarcoidosis (clinical and radiological observation)

Anatoliy V. Lenshin, Andrey V. Ilin, Elena A. Ignatieva, Juliy M. Perelman*

Abstract

Bronchostenosis is one of relatively rare complications of thoracic sarcoidosis. We have retrospectively analyzed the results of comprehensive examinations, including DICOM images, of a cohort of 100 patients with confirmed sarcoidosis who were in the clinic and examined on an outpatient basis at the Far Eastern Scientific Center of Physiology and Pathology of Respiration (Blagoveshchensk, Russia) in 2018-2019. All of them underwent, along with clinical examination, multislice computed tomography (MSCT) with basic software post processing: multiplanar two projection reforming (MPR), three dimensional rendering (3DR), virtual bronchoscopy (VBS), etc. The obtained MSCT results were compared with fibrobronchoscopy (FBS) and transbronchial biopsy under ultrasound guidance. We report a case of a 52 year old woman with stenosis of the right middle lobe bronchus with atelectasis of the middle lobe of the right lung due to sarcoidosis. She was examined on an outpatient basis by multislice computed tomography with post processing. MSCT with postprocessing makes it possible to: study the stenotic area along the entire length (inaccessible to FBS in a significant degree of stenosis); studying the structure of the bronchial wall (thickness, density, size of lesions); analysis of endobronchial inclusions, study of extrabronchial tissue structures (compressing ITLN, etc.). The use of the full potential function of modern MSCT technologies creates an opportunity to carry out expert non-invasive diagnostics of various manifestations of sarcoidosis, even in primary health care.

Keywords: Sarcoidosis; Bronchostenosis; Atelectasis; Multispiral computed tomography; Postprocessing

Introduction

Sarcoidosis is a multisystem granulomatous disease of unknown etiology. The dominant (more than 90%) localization is the organs of the thoracic cavity with damage to the mediastinal lymph nodes and lung parenchyma. The airways are damaged significantly less often as a result of both endobronchial granulomatous inflammation and as a result of external influence (compression) by enlarged intrathoracic lymph nodes (ITLN),

especially with their calcification.¹ Airway involvement may lead to airflow limitation with hypoventilation and even atelectasis of the segment or the entire lobe.^{1,2} In the historical aspect, it should be emphasized that the first case of endobronchial sarcoidosis was described by E.B. Benedict, B. Castleman back in 1941.³ Endobronchial lesion complicated by stenosis of the central or proximal bronchi, and sometimes even trachea⁴, accompanied by segmental or lobar atelectasis is a potentially severe manifestation of sarcoidosis. Unusual (atypical) clinical presentation and variable response to medical treatment require specific attention to diagnosis and follow up.² Although the classic forms of sarcoidosis with ITLN involvement are familiar to most clinicians and radiologists, airway involvement is often missed due to information deficiency and technological imperfections. Possibilities of modern methods of radiological diagnosis of this pathology, including high resolution computed tomography (HRCT), are insufficiently covered in literature. We report a case of a 52 year old woman with stenosis of the right middle lobe bronchus due to sarcoidosis.

Materials and methods

We have retrospectively analyzed the results of comprehensive examinations, including DICOM images, of a cohort of 100 patients with confirmed sarcoidosis who were in the clinic and examined on an outpatient basis at the Far Eastern Scientific Center of Physiology and Pathology of Respiration (Blagoveshchensk, Russia) in 2018-2019. All of them underwent, along with clinical examination, multislice computed tomography (MSCT) with basic software post processing: multiplanar two projection reforming (MPR), three dimensional rendering (3DR), virtual bronchoscopy (VBS), etc. The obtained MSCT results were compared with fibrobronchoscopy (FBS) and transbronchial biopsy under ultrasound guidance.

Results

9 patients with bronchial stenosis (9%) were identified. The median age was 49 years, with a range of 17-64 years. All patients were female. Bronchial stenosis was caused by external compression of ITLN in 4 patients, by endobronchial sarcoidosis in 3 patients, and by a combination of ITLN compression and endobronchial sarcoid lesion in 2 patients.

Own clinical and radiological examination of one of the observations

Patient M, 52 years old. At the time of the study, she complained of a scanty cough, shortness of breath when climbing up to the 3rd floor. Radiographs were taken twice, 6 months apart. Pneumonia in the middle lobe of the right lung with a protracted course was diagnosed, because the X ray pattern remains stable despite the ongoing therapy. The patient was referred for further diagnosis in the Far Eastern Scientific Center of Physiology and Pathology of Respiration. MSCT was performed (Figure 1). The conclusion was drawn pulmonary mediastinal form of sarcoidosis of the stage 2, stenosis of the right middle lobe bronchus, atelectasis of the mid-

Laboratory of Functional Research of Respiratory System, Far Eastern Scientific Center of Physiology and Pathology of Respiration, Blagoveshchensk, Russia

*Corresponding author: Juliy M. Perelman
e-mail: jperelman@mail.ru*

Case Report

dle lobe. Stenosis is due to external compression of the ITLN and endobronchial sarcoid inflammation.

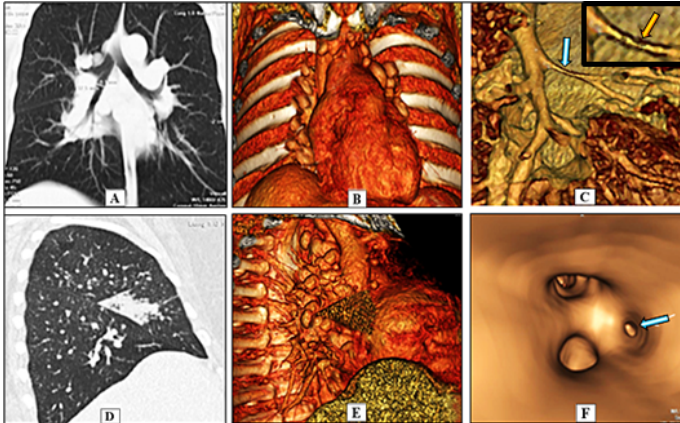


Figure 1: Patient M, 52. Pulmonary-mediastinal form of sarcoidosis, stage 2, stenosis of the right middle lobe bronchus with atelectasis of the middle lobe. MSCT.

Radiological features

The figure shows a logical combination of fragments of various reconstructions of the MSCT study of patient M, each of which carries its own significant value of image information. Fragment (A) MPR in coronal plane a pronounced bilateral increase in ITLN is determined. (B) 3DR in the coronal plane bilateral, mainly hilar, increase in ITLN. (C) 3DR in the right lateral plane circular narrowing of the right middle lobe bronchus (indicated by an arrow), in the upper right corner of this fragment a localized enlarged section of the narrowing zone is shown thickened, compacted walls of the bronchus are visualized, and in the lumen there are granulomatous (polypous?) focal consolidations (indicated by a yellow arrow), emphasizing the degree of stenosis. (D) MPR in the right lateral plane a characteristic, triangular shaped radiological pattern of atelectasis of the middle lobe of the right lung is determined. (E) 3DR in the right lateral plane a triangular shaped atelectasis of the middle lobe is visualized, surrounded by compressing ITLNs. (F) VBS of the right lung a sharp, up to 2 mm-3 mm, circular narrowing of the middle lobe bronchus (indicated by the arrow).

Fibrobronchoscopy

A sharp narrowing of the lumen of the right middle lobe bronchus, mucous membrane swelling, erythematous areas, granularity and looseness of the mucous membrane, granulomatous sarcoid tubercles like “cobblestone pavement”.

Spirometry

Mild airflow limitation.

Discussion

Airway sarcoidosis with bronchial stenosis and even more so atelectasis is a rare form in the cohort of atypical sarcoidosis that requires correction of diagnosis and therapy. Its prevalence among verified sarcoidosis varies widely: from 0.72% when performing HRCT 2 up to 14% when performing FBS 5 and even up to 26%.⁶ These statistics indicate that radiological methods, including HRCT, have not fully demonstrated their potential in the diagnosis of atypical forms of sarcoidosis, and this dictates the need for further improvement of methods for visualizing this pathology.

By all means, the diagnosis of bronchial stenosis in sarcoidosis should be complex, using, in addition to clinical assessment, FBS with biopsy, MSCT with postprocessing, and methods for lung function assessment. The specific contribution of the MSCT study is shown in the figure in our clinical and radiological observation. MSCT with postprocessing makes it possible to study the stenotic area along the entire length (inaccessible to FBS in a significant degree of stenosis); to study the structure of the bronchial wall (thickness, density, size of lesions); to analyze the endobronchial inclusions; to study the extrabronchial tissue structures (compressing ITLN, etc.).

Conclusion

The use of the full potential function of modern MSCT technologies creates an opportunity to carry out expert non-invasive diagnostics of various manifestations of sarcoidosis, even in primary health care.

References

1. Hunninghake GW, Costabel U, Ando M, et al. ATS/ERS/WASOG statement on sarcoidosis. American Thoracic Society/European Respiratory Society/World Association of Sarcoidosis and other Granulomatous Disorders. *Sarcoidosis Vasc Diffuse Lung Dis* 1999; 16(2):149-173.
2. Chambellan A, Turbie P, Nunes H, et al. Endoluminal stenosis of proximal bronchi in sarcoidosis: Bronchoscopy, function, and evolution. *Chest* 2005; 127(2):472-481.
3. Benedict EB, Castleman B. Sarcoidosis with bronchial involvement. *N Engl J Med* 1941; 224:186-189.
4. Ulmeanu R, Răjnoveanu A, Halic E, et al. Endobronchial changes in sarcoidosis. *Pneumologia* 2011; 60(3):155-159.
5. Stjernberg N, Thunell M. Pulmonary function in patients with endobronchial sarcoidosis. *Acta Med Scand* 1984; 215(2):121-126.
6. Armstrong JR, Radke JR, Kvale PA, et al. Endoscopic findings in sarcoidosis. Characteristics and correlations with radiographic staging and bronchial mucosal biopsy yield. *Ann Otol Rhinol Laryngol* 1981; 90(4, Pt 1):339-343.