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Mysterious plastic bronchitis: a little-known disease in medical practice

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ABSTRACT

The article presents a clinical case of a 51-year-old patient first seeking medical care with complaints of paroxysmal cough bringing up bronchial casts. The diagnosis of plastic bronchitis was verified. The disease which has not been well described in the literature, difficulty of verifying the underlying diagnosis due to polysymptomatic clinical presentation characterized by the mortality rate of 50–80%, COVID-19 coinfection, resistance to therapy, and little concern of medical specialists determine the relevance and value of this clinical case.

Keywords: bronchial casts, plastic bronchitis, lymphatic plastic bronchitis, bronchial obstruction syndrome

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Загадочные «резиновые» слепки бронхов: малоизвестное заболевание в медицинской практике

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РЕЗЮМЕ

Представлено клиническое наблюдение пациентки, впервые обратившейся за медицинской помощью в возрасте 51 года по поводу приступообразного кашля с отхождением слепков бронхов. Верифицирован диагноз лимфопластического бронхита. Нозология, ранее не нашедшая подробного отражения в литературе, трудность верификации основного диагноза на фоне полисимптомной клинической картины, характеризующейся летальностью в 50–80%, сочетанное течение с новой коронавирусной инфекцией, а также резистентность к проводимой терапии и низкая настороженность медицинского сообщества определяют актуальность и ценность данного клинического наблюдения.

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Ключевые слова: слепки бронхов, пластический бронхит, лимфопластический бронхит, бронхообструктивный синдром

Конфликт интересов. Авторы декларируют отсутствие явных и потенциальных конфликтов интересов, связанных с публикацией настоящей статьи.

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INTRODUCTION

Increased interest in respiratory pathology in recent years is explained by the pandemic of the SARS-CoV-2 coronavirus. However, bronchial obstruction syndrome in adults can also be caused by rare nosological entities, which include plastic bronchitis.

Plastic bronchitis (PB) (chronic fibrinous bronchitis, pseudomembranous bronchitis, mucoid impaction, Hoffman's bronchitis, lymphatic (lymphoid, lymphatic plastic) bronchitis) is a chronic recurrent bronchial inflammation, characterized by the formation of rubbery plugs (in the form of bronchial casts that are denser than normal mucous secretions) causing airway obstruction and respiratory failure [1–7].

The first descriptions of bronchial casts date back to the time of Galen (131–200 AD) who characterized them as "expectorated arteries and veins" [4, 8]. In 1750, Bussiere reported a postmortem examination of a patient suffering from tuberculosis. The patient was also found to have a bronchial cast *in situ* [4]. In 1902, M. Bettman described several cases of PB with bronchial casts and R. Shaw documented cases of mucoid impaction in 1951 [8]. By 1960, about 300 cases had been described in the literature [9, 10]. About 420 PB cases had been registered by 2008 [2, 10].

When searching for the term "plastic bronchitis" in the English-language text database PubMed in 2022, one could find 513 publications. Meanwhile, a detailed search for "adult plastic bronchitis" showed only one-fourth of these publications – 127. From 1965 to 2022, the number of manuscripts devoted to this topic increased, with the largest number of articles, reviews, and cases published in the last decade.

In our country, there are only few publications describing such clinical cases so far. Most of them, as well as in the foreign literature, are related to pediatrics. This can be explained by the peculiarities of the disease. It is diagnosed more often in children and at a young age. In adults, the disease is registered very rarely [1–6, 8, 9]. These cases in the absence of infectious and allergic genesis can be called casuistic. It is more common in women than in men [5]. At the same time, there is an opinion that it is slightly more frequent in men [4]. A higher prevalence is registered in dry and hot areas due to the changes in physical and chemical properties of airway secretions as a result of mucosal dehydration [10].

Unfortunately, there have been no large-scale epidemiological studies of PB in adults. The true incidence and prevalence rates cannot be accurately determined because it is likely that many episodes of the disease are left undiagnosed. The largest pool of observations of lymphatic PB in adults (n = 44) was published in 2022 by C. O'Leary et al. [11]. In Russia, the observational data of the largest cohort of PB patients (n = 20) for the period from 1990 to 2012 were published by V. Molodtsova et al. [5]. In 2021, Japanese scientists Y. Murata et al. described a case of the oldest patient. The 74-year-old woman had multiple myeloma and PB associated with a viral infection (influenza A virus) [11]. The problem is highly alarming not only due to underdiagnosis of this condition, but also because of a high mortality rate, which reaches 50–80%, according to some authors [4, 6, 7, 9].

The etiology and pathogenesis of the disease still remain completely uncertain (Table).

The clinical presentation of PB includes cough, dyspnea, crackles in the lungs, and cough bringing up bronchial casts. The course of the disease is complicated by atelectasis, bronchial obstruction syndrome, and fatal asphyxia [1, 3, 4].

Table

Etiology of plastic bronchitis*	
Etiology	Associated diseases
Primary and secondary lymphatic abnormalities	Congenital heart failure (heart defects) following surgical correction (Fontan, Glenn, Blalock – Taussig procedures). Chest trauma. Lymphatic abnormalities, lymphangiectasia, lymphangiomatosis
Viral infections	Influenza A virus, adenovirus infection, rhinovirus infection, respiratory syncytial virus, parainfluenza virus, SARS-CoV-2
Other lung diseases	Bronchial asthma, bronchopulmonary aspergillosis, bronchiectasis, cystic fibrosis
Bacterial infections	Streptococci, Haemophilus influenzae, Mycobacterium tuberculosis, Klebsiella, etc.
Hematological diseases	Acute respiratory distress syndrome in sickle cell anemia
Occupational diseases	Silicosis
Cancer	Kaposi's sarcoma with possible involvement of lymphatic vessels. Endobronchial metastases in kidney cancer

*Adapted from [4], [13].

The diagnosis of PB is confirmed by the presence of casts that have been coughed up or visualized during fibrobronchoscopy (FBS). A full clinical and laboratory examination is required for diagnosis [10]. The gold standard for verification and treatment of PB is dynamic contrast-enhanced magnetic resonance lymphangiography and intranodal lymphangiography, lymphatic embolization [10, 11] (Fig. 1).

CLINICAL CASE

Female patient V., 51 years old, born in Kogalym, is a doctor herself. The patient first sought medical help in April 2020, complained of paroxysmal cough bringing up bronchial casts. The casts had dense consistency (rubbery), white color (Fig. 2) and were coughed up mainly at night and in the morning, sometimes accompanied by abundant liquid sputum of milky white color. "Nagging" cough, "choking" with sputum at night, increased body temperature, shortness of breath, and difficulty exhaling during normal physical activity.

The patient became acutely ill in March 2020, when the clinical symptoms of acute respiratory viral infection appeared (body temperature increased to 38 ºC, runny nose, paroxysmal cough). The therapy (with antiviral drugs, expectorants, mucolytics) resulted in few positive changes: cough improved, body temperature lowered, rhinorrhea disappeared.

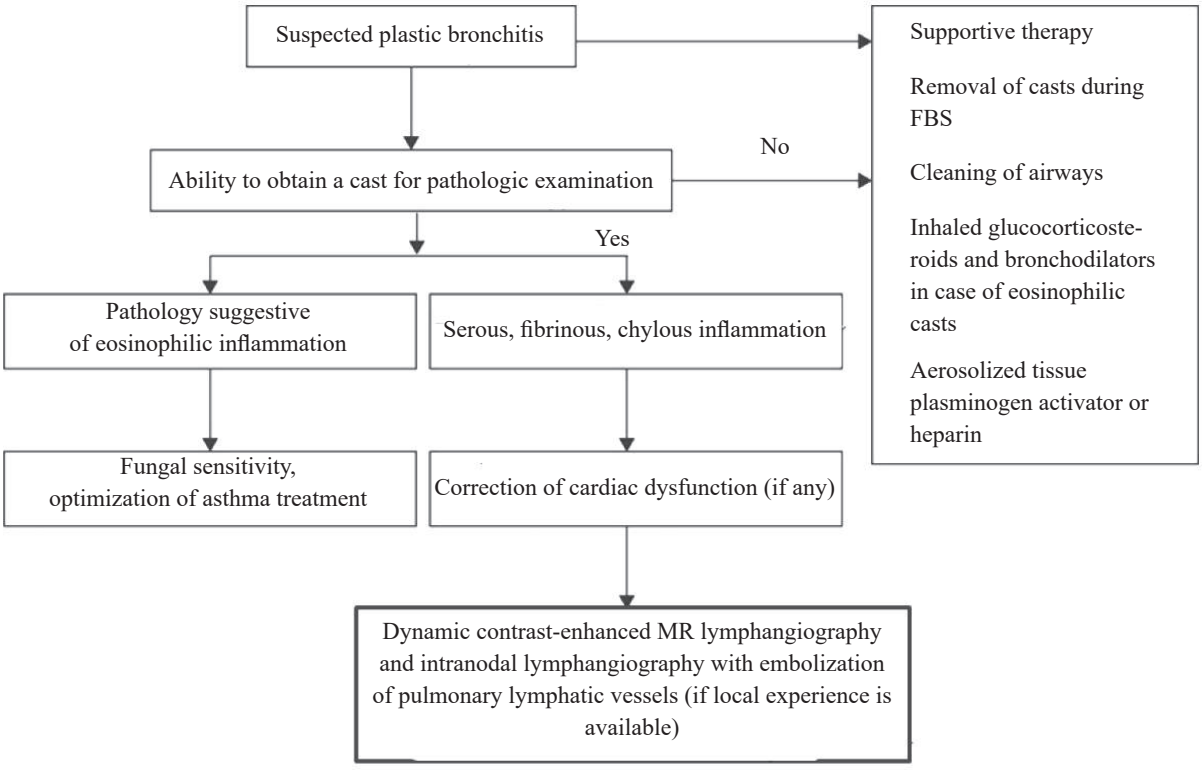


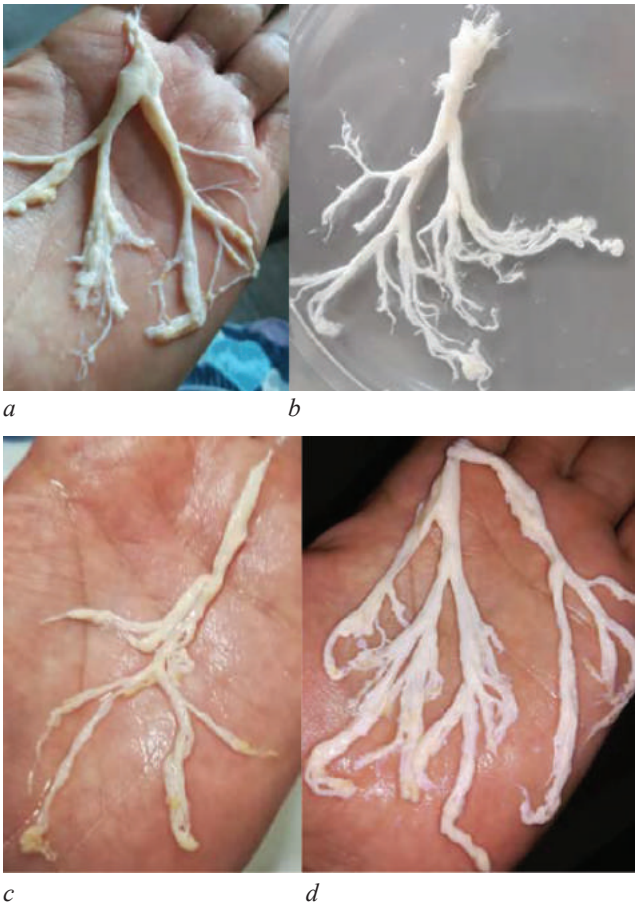
Fig. 1. Algorithm of diagnosis and management of PB patients [4]

Two weeks later, the patient first complained of abundant expectoration of milky-white sputum. For the first time, she coughed up small rubbery bronchial casts. Past medical history: hypertension and type 2 diabetes mellitus verified 5 years ago. In 2010, she underwent left mastectomy for breast cancer. Objective examination findings: postoperative scar in the area of the left breast, no abnormality detected, body mass index 33.3 kg / m^2, rough breathing on the left side in the lower parts of the lungs, and oxygen saturation of 96%.

Based on the available data, the patient was diagnosed with acute bronchitis. The antibacterial therapy that was prescribed did not have any effect. A month after a contact with a COVID-19 patient, a woman reported that her condition aggravated: body temperature increased to 39ºC, cough became stronger, the volume of sputum increased, dyspnea appeared. The patient was diagnosed with COVID-19. Doctors made a differential diagnosis with pneumonias of different infectious genesis, tuberculosis, and allergic lung damage. The FBS findings were remarkable: on the left, the upper lobe bronchus orifice was obturated with cheesy necrotic mass, mucosa was hyperemic, the lumen contained a moderate amount of turbid sputum. The patient received treatment according to the temporary clinical guidelines for COVID-19, as well as symptomatic therapy. During the FBS, the patient's necrotic tissue was removed and airways were cleared. After discharge, the patient's condition rapidly deteriorated: dyspnea became worse, cough became hacking and more intense, the volume of expectorated sputum increased. The patient started to cough up bronchial casts again at night and in the morning (Fig. 2).

Each paroxysm ended with coughing up a cast. Due to the deterioration of symptoms, the patient was referred to the Ural Research Institute of Phthisiopulmonology (23.06.2022–13.07.2022).

Lung computed tomography (CT) revealed bilateral polysegmental lung lesion in the regression phase, left lung lesion – ground glass opacity, reactive lymphadenopathy of intrathoracic lymph nodes. A cytological examination of the casts was performed: mucus clumps with groups of bronchial epithelial cells having reactive cellular changes, few macrophages with leukocytes (bronchial casts), admixture of traumatic tap blood. There was no data indicating tumor growth (Fig. 3). The available data suggested the diagnosis of PB.



a *b* *c* *d*

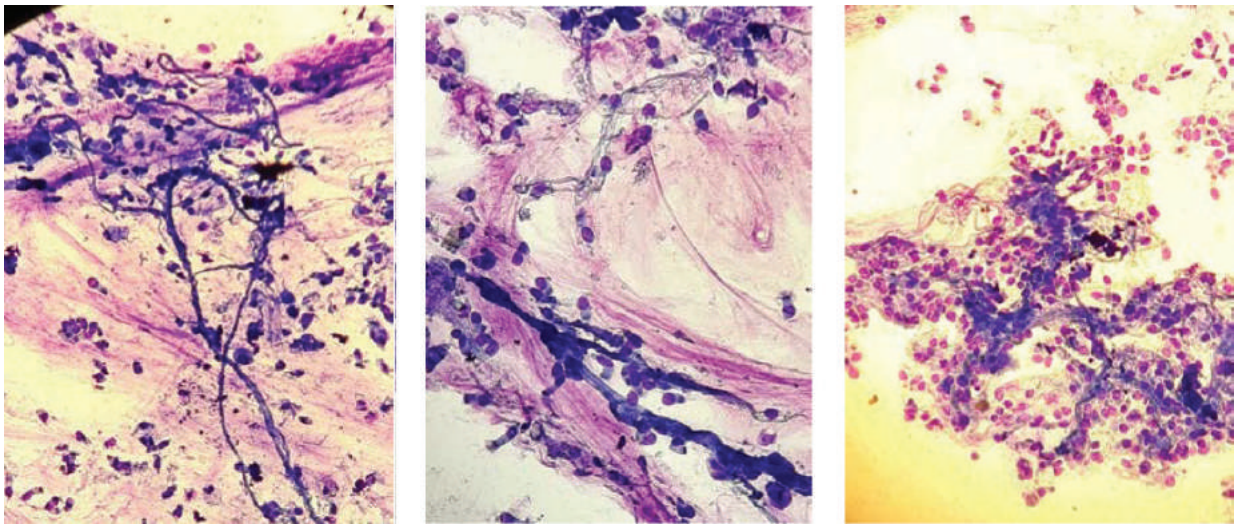
Fig.2. Patient V., 51 years old. Sputum in the form of bronchial casts

A woman was consulted by V. Parshin, thoracic surgeon, Doctor of Medical Sciences, Professor, corresponding member of the Russian Academy of Sciences, Head of the Surgical Department of the I.M. Sechenov Clinical Center. The patient was also remotely consulted by Maxim Itkin, thoracic surgeon, Professor of Radiology at the Hospital of the University of Pennsylvania, Director of the Center for Lymphatic Imaging and Interventions (Philadelphia, USA). Based on the features that can be considered the disease criteria, which are: ground glass opacity of one lung according to lung CT, sputum in the form of bronchial casts, lymphadenopathy of the intrathoracic bronchial nodes, obesity, increased expectoration of sputum after eating fatty foods, absence of convincing data about an allergic process, PB of suspected lymphatic genesis was confirmed. The detection of neutral fat in bronchial lavage fluid would have been an additional criterion, but this procedure was not carried out.

In February 2021, doctors performed ligation of the thoracic duct above the diaphragm and transection of

the lymphatic vessels of the left lung root according to the root skeletonization method at the Department of Thoracic Surgery of the Sechenov University Clinical Hospital No.1.

Thirty minutes prior to the surgical intervention, the patient was offered to drink 200 ml of a fatty mixture (sour cream (20%), cream (10%), and butter) to detect a lymphatic vessel leakage. After that, the chest was opened and a lymphatic vessel causing abundant lymphorrhea was found. A cyst of the thoracic lymphatic duct was also detected intraoperatively. In the early postoperative period, the patient started to adhere to an oral diet of fat-free food gradually extending a range of food products. Final diagnosis: thoracic duct cyst (parabronchial, collateral cyst on the left). Lymphatic plastic bronchitis (chylobronchorrhea). Lymphadenopathy of the mediastinum. Recommendations: low-fat diet (1 month), expanding meals after 1–1.5 months, restriction of physical activity for 2 months, correction of body weight. One year later, the woman had no complaints, the above symptoms were eliminated completely, the patient did not follow the diet. According to the lung CT (April 2022), the patient still had pulmonary fibrosis of the left lung.



a *b* *c*

Fig. 3. Cytological examination of bronchial casts of patient V

DISCUSSION

Difficulties making the diagnosis in this case were associated with the clinical polymorphism of the disease, low awareness of specialists about this pathology, COVID-19 coinfection, which could have both initiated and masked the disease, and the lack of convincing data during the examination for a lymphatic vessel defect causing the patient's symptoms.

The true etiology of the pathological changes in the observed woman remains an open question. Several causes and factors may be involved. In recent years, some scientists have supposed that abnormal communication and leakage of lymphatic fluid into the airway are some of the main PB causes. Lymphatic duct obstruction can explain PB in cardiac surgery, heart failure or trauma [9, 12]. Many cases of idiopathic PB in adults are associated with lymph, which allows us to rename the diagnosis in these patients into "lymphatic plastic bronchitis (lymphatic PB)" [7, 11]. It is possible that the left mastectomy performed 12 years ago caused pathological changes in this patient. Increased pressure in the lymphatic vessels observed in congenital lymphangiectasia or lymphangiomatosis can lead to retrograde lymph flow, drainage of lymph into the bronchial lumen, and chylothorax development [13].

CONCLUSION

The disease has not been well described in the literature. It is difficult to diagnose due to the poly-symptomatic clinical presentation and manifestations that coincide with different bronchopulmonary pathologies. The disease is characterized by the mortality rate of 50–80%, COVID-19 coinfection, resistance to therapy, and low awareness of medical

specialists about it. These reasons determine the relevance and value of this clinical case.

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