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Длительный анамнез бронхоцеле, вызванный типичным карциноидом

К.В. Прусакова, П.В. Гаврилов

Санкт-Петербургский научно-исследовательский институт фтизиопульмонологии, Санкт-Петербург, Российская Федерация

АННОТАЦИЯ

В работе представлен клинический случай с длительным периодом наблюдения одиночного бронхоцеле (бронхогенной ретенционной кисты). При первоначальном комплексном обследовании, включающем такие исследования, как рентгенография, компьютерная томография органов грудной полости, фибробронхоскопия, иммунологические и бактериологические обследования на туберкулёз, данных за онкологическую и инфекционную природу изменений не выявлено. Изменения были расценены как последствия перенесённого неспецифического воспалительного процесса. Через 15 лет при плановом медицинском осмотре по данным рентгенографии органов грудной полости отмечено увеличение размеров бронхоцеле, а также появление округлого образования в медиальных отделах бронхоцеле. С помощью дополнительных методов исследования, таких как компьютерная томография органов грудной полости с внутривенным контрастированием, фибробронхоскопия с биопсией, установлено, что выявленное образование является типичным карциноидом.

Несмотря на то что бронхоцеле в большинстве случаев является доброкачественным изменением, из разнообразия причин, вызывающих его развитие, следует выделить обструкцию бронха новообразованием. Среди новообразований лёгкого типичный карциноид составляет всего 1–2%, характеризуется крайне медленным ростом и отсутствием специфичной клинической симптоматики. Несмотря на это, типичный карциноид относится к злокачественным нейроэндокринным образованиям I типа. В 10–15% случаев выявляются метастазы, преимущественно в медиастинальные лимфатические узлы, а также в печень, кости, реже в мягкие ткани.

Данное клиническое наблюдение говорит о том, что даже при отрицательных результатах первичного обследования локально расположенного бронхоцеле такие изменения требуют онкологической настороженности и периодических обследований в динамике.

Ключевые слова: клинический случай; бронхоцеле; типичный карциноид; компьютерная томография.

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Long-term broncocele anamnesis, triggered by typical carcinoid

Kseniya V. Prusakova, Pavel V. Gavrilov

Saint-Petersburg State Research Institute of Phthisiopulmonology, Saint Petersburg, Russian Federation

ABSTRACT

The paper presents a case of a single bronchocele (bronchogenic retention cyst) caused by a typical carcinoid that was observed for a long time. During the initial complex examination, including computed tomography with intravenous contrast, fibrobronchoscopy, and immunological and bacteriological examinations of tuberculosis, there were no changes for the oncological and infectious nature. The changes were interpreted as the result of a postponed nonspecific inflammatory process. Most of them were monitored using chest X-ray and the changes were stable. After 15 years, a control chest X-ray revealed an increase in the size of the compaction in the lung and the appearance of a mass with calcification in the medial sections of the compaction zone. Additional examination, including computed tomography with biopsy, determined that the obstruction of the bronchus was caused by a neoplasm [according to histological examination (typical carcinoid)].

It should be noted that the initial detection of negative study results requires oncological alertness and periodic examinations in dynamics.

Keywords: case report; bronchocele; typical carcinoid; computed tomography.

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由典型的类癌引起的支气管囊肿的悠久历史

Kseniya V. Prusakova, Pavel V. Gavrilov

Saint-Petersburg State Research Institute of Phthisiopulmonology, Saint Petersburg, Russian Federation

简评

本文提出了一个长期观察单个支气管囊肿（支气管原性保留囊肿）的临床病例。在最初的全面检查中，包括放射照相术，胸腔计算机断层扫描，纤维支镜检查，结核病的免疫学和细菌学检查等研究，没有发现改变的肿瘤和感染性的数据。这些变化被视为转移非特异性炎症过程的后果。15年后，在常规体检期间，根据胸腔的射线照相，注意到支气管的大小增加，以及支气管囊肿内侧部分圆形出现。在其他研究方法的帮助下(例如胸腔静脉造影的计算机断层扫描，活检的纤维支镜检查)，确定检测到的形成是典型的类癌。

尽管在大多数情况下，支气管囊肿是一种良性变化，但从导致其发展的各种原因来看，有必要通过肿瘤来区分支气管囊肿的阻塞。在肺部肿瘤中，典型的类癌仅为1-2%，其特征在于极其缓慢的生长和没有特定的临床症状。尽管如此，典型的类癌属于第一类型恶性神经内分泌形成。在10-15%的病例中，检测到转移，主要在纵隔淋巴结中，以及在肝脏，骨骼中，在软组织中较少。

这一临床观察表明，即使对局部定位的支支气管囊肿的初步检查结果为阴性，这种变化也需要肿瘤警觉性和动态的定期检查。

关键词： 临床病例；支气管囊肿；典型类癌；计算机断层扫描。

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INTRODUCTION

Bronchocele (bronchogenic retention cyst, mucocele) is a relatively common finding in chest X-ray studies. The morphological substrate of bronchocele is local bronchiectasis in which airways are filled with mucous contents persistently secreted by the mucous membrane and with proximal obstruction of the airways [1]. In radiography and computed tomography, bronchocele is visualized as a tubular branched V- or Y-shaped structure associated with the bronchial tree (finger in glove sign) [2]. The contents have homogeneous structures, but dense inclusions (calcifications) are visualized in 30% of the cases [2, 3]. The contrast agent is not accumulated in computed tomography with intravenous contrast enhancement.

Bronchocele can have an oval or round shape, which depends on the size of the obturated bronchus, amount of contents in the lumen, and state of the surrounding pulmonary parenchyma.

Solitary local retention cysts are asymptomatic. Retention cysts have various causes, such as congenital diseases (bronchial atresia, lung sequestration, and cystic fibrosis), infectious pathologies (nonspecific inflammatory processes, tuberculosis, mycobacteriosis, and allergic bronchopulmonary aspergillosis), obstruction of the bronchus by the lesion (benign or malignant), foreign body, or cicatricial deformity of the bronchus. Differential diagnostics is complicated because bronchocele can have similar radiological semiotics regardless of causes [2].

Bronchocele should be differentiated with arteriovenous malformations in the lungs, such as endobronchial metastasis. In this case, computed tomography with intravenous contrast enhancement is the preferred diagnostic method [2].

In most cases, bronchocele is caused by benign changes in the lungs and does not require case follow-up; however, in a locally located bronchocele, obstructive genesis by the lesion or foreign body should be ruled out. For this purpose, supplementing radiation diagnostic methods with fibrobronchoscopy with biopsy is recommended [4, 5].

Currently, an optimal diagnostic algorithm for identifying the cause of bronchocele development has not been established. Moreover, there are no uniform recommendations for further follow-up of patients with newly diagnosed asymptomatic retention cysts or bronchocele.

CASE DESCRIPTION

A 56-year-old male patient visited the Department of Radiation Diagnostics for computed tomography of the chest cavity organs.

The history assessment revealed that he was examined for pneumonia 15 years ago. Despite the positive dynamics based on clinical studies, during the course of antibiotic therapy, radiological findings did not correspond to the typical course of regression of infiltrative lungs changes in pneumonia. X-ray imaging of the chest revealed an area of induration of a tubular branched structure in the middle section of the right lung (Fig. 1, *a*). Additional studies, including computed tomography of the chest with intravenous contrast enhancement, fibrobronchoscopy, and immunological and bacteriological studies, did not detect tuberculosis or an oncological process. Computed tomography data were presented as selective scans on a film carrier, which revealed a local, single branched structure with smooth, clear contours, located along the subsegmental bronchi of the middle lobe of the right lung (finger in glove sign), with homogeneous contents (Fig. 2), so the patient was diagnosed with bronchogenic retention cyst (bronchocele) on the middle lobe of the right lung. Subsequently, follow-up studies were performed annually by X-ray examination of the chest, and stable changes were observed.

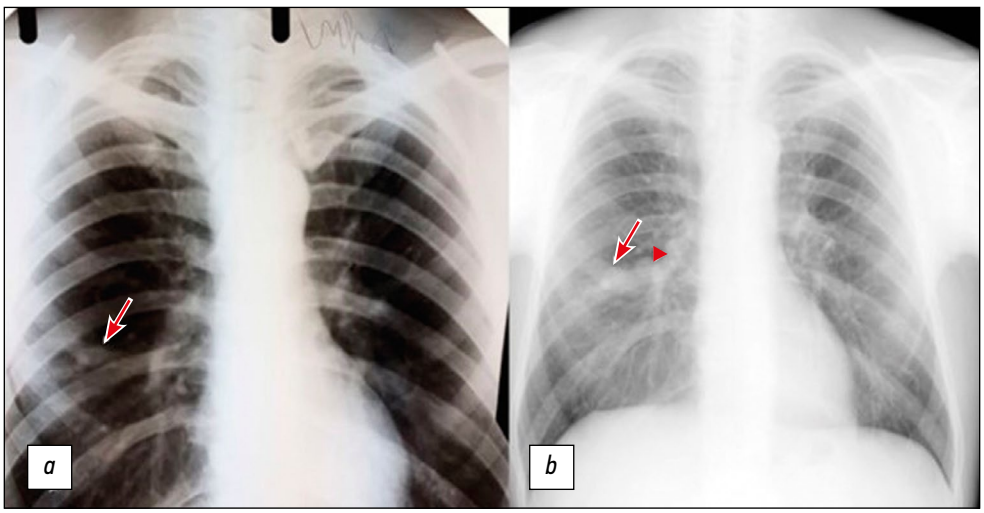

Fig. 1. X-ray image of the chest cavity organs of a 56-year-old patient *a,* At age 41 years, initial examination of the middle section of the right lung revealed a segment of induration of the branched tubular structure (arrow); *b,* 15 years later, the size of the bronchocele (arrow) increased, and a rounded lesion in the medial parts of the bronchocele (arrowhead) emerged.

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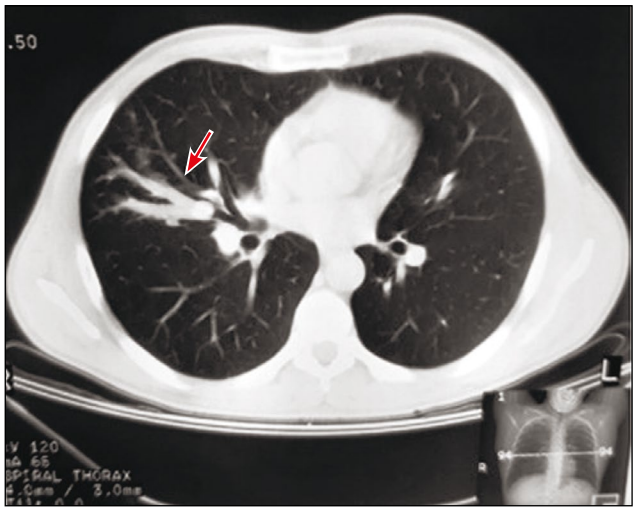

Fig. 2. Selective computed tomography scan of the thoracic cavity organs of the same patient: a homogeneous V-shaped structure in the middle lobe of the right lung with clear contours (arrow).

Prior to the present admission, the patient underwent a medical examination at his workplace with harmful working conditions. X-ray imaging of the chest revealed an increase in the size of the previously determined bronchocele (Fig. 1, *b*), as well as a new round lesion in the medial sections of the bronchocele with calcifications along the lesion contour (Fig. 1, *b*). To clarify the nature of the changes, the patient underwent contrast-enhanced computed tomography of the chest, which detected a single branched V-shaped structure with a clear contour in the middle lobe of the right lung, and homogeneous contents located along the subsegmental bronchi (finger in glove sign) were preserved. At the base of the bronchocele, a rounded lesion with a smooth, clear contour is noted, almost completely overlapping the bronchus B4 lumen, and single calcifications were found along the periphery with signs of contrast accumulation in the venous phase from +29 HU to +112 HU (Fig. 3). Changes were characteristic of bronchocele caused by neoplastic bronchus obstruction. Fibrobronchoscopy with biopsy was also performed. Bronchoscopy revealed a rounded lesion of the B4 ostium, which completely covered the bronchial lumen (Fig. 4). The lesion is inactive and woundable on contact, and the mucous membrane on the surface is hyperemic and edematous. The biopsy results revealed that the histological presentation of the lesion corresponded to a typical carcinoid. The immunohistochemical study revealed that tumor cells intensely expressed CD56, but not TTF1. The Ki67 proliferative activity index was 2%.

The patient received surgical treatment by resection of the middle lobe of the right lung. On 1-year follow-up examination, no signs of carcinoid recurrence were observed by computed tomography of the chest.

DISCUSSION

The most common causes of multiple bronchocele formation are cystic fibrosis, allergic bronchopulmonary

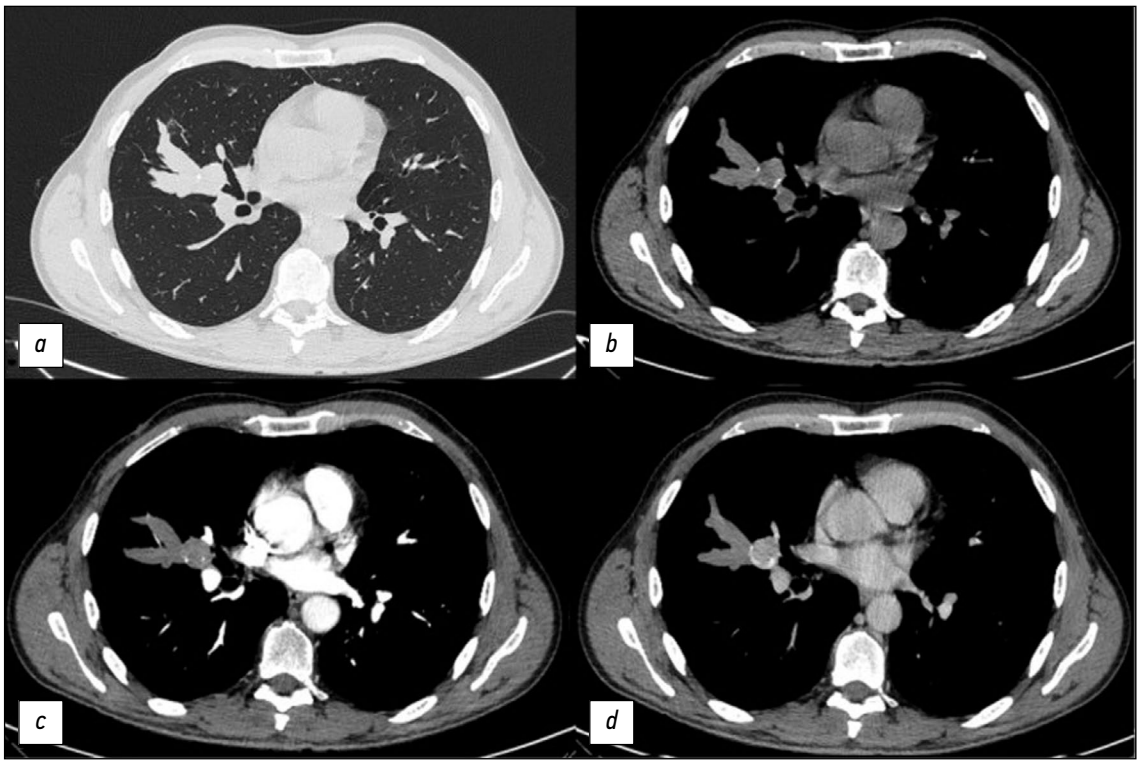

Fig. 3. Computed tomography scan of the chest cavity organs in the axial plane in the same patient: *a,* lung window; round lesion at the base of the bronchocele was detected during the native phase; *b,* mediastinum window; single calcifications along the periphery of the lesion were noted; *c,* mediastinum window; arterial phase; *d,* mediastinum window; signs of contrast accumulation by the lesion were detected in the venous phase.

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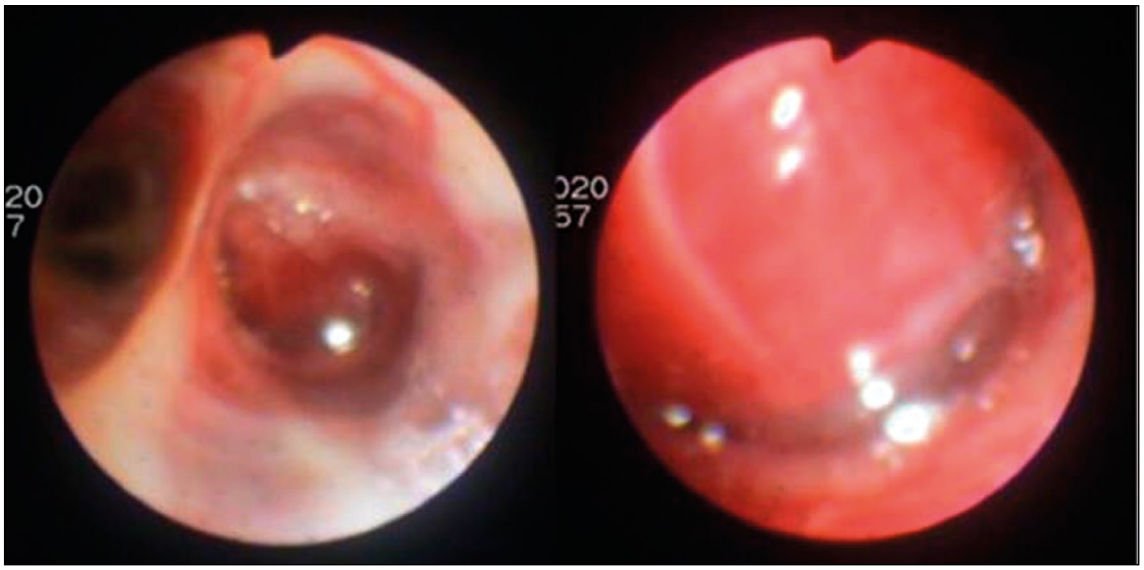

Fig. 4. Fibrobronchoscopy in the same patient. The lesion of the B4 ostium on the right completely blocked the bronchus lumen.

aspergillosis, and tuberculosis. Solitary local retention cysts are more often caused by the obstruction of the bronchus by a neoplasm (benign or malignant) [2, 6].

A typical carcinoid accounts for 1%–2% of lung neoplasms [7]. In 70% of the cases, the tumor is localized in the main bronchi, more often in the right lung, primarily in the middle lobe [8]. Typical carcinoid is commonly observed in people aged 40–50 years. With this form of lung neoplasm, studies have not established a reliable relationship between carcinogens and smoking [9, 10].

In most cases, bronchial carcinoid is asymptomatic and is detected as an accidental finding during a routine examination; however, in 2%–5% of the cases, bronchial carcinoids can produce neuroamines and peptide hormones, such as serotonin, adrenocorticotropic hormone, somatostatin, and bradykinin [11]. Clinical manifestations of carcinoid syndrome include periodic hot flashes or a sensation of blood rushing to the head, neck, and arms, bronchospasm, diarrhea, and mental disorders [11–13].

On X-ray imaging, a typical carcinoid is seen as a round or oval lesion with clear and even (sometimes lobular) contours. In up to 30% of the cases, eccentrically located or diffuse calcifications are observed [2, 3].

On computed tomography, a typical carcinoid is revealed as a rounded lesion with clear, even, or lobed contours. With intravenous contrast enhancement, there is an accumulation of a contrast agent, and in some cases, it is possible to trace the feeding artery entering the lesion from the bronchial arteries [6]. In relation to the bronchus, the carcinoid was located intrabronchially, extrabronchially, or mixed iceberg type, causing partial or total obstruction of the bronchial lumen [2, 3].

In the present case, although the cause of the bronchocele development was not established in the initial comprehensive examination, retrospective assessment of computed tomography data presented on a film carrier revealed the presence of a lesion at the base of the bronchocele (Fig. 2). With its extrabronchial location, changes during fibrobronchoscopy may not be detected.

The densitometric parameters of the lesion located at the base of the retention cyst may not be substantially different from the mucus, and small ones can be difficult to visualize. Central carcinoid may be suspected when signs of obstruction (atelectasis, "air traps," or bronchocele) are detected.

Differentiation of a typical carcinoid should be performed with type II neuroendocrine lesions of the lungs (atypical carcinoid), bronchogenic cyst, and bronchocele.

The typical carcinoid is extremely slow growing. According to Raz et al. [14], the average doubling time of typical carcinoid tumors is 7 years; therefore, it is difficult to judge the dynamics based on the annual prophylactic radiography of the lungs, since it is difficult to detect visually a minor increase in tumor size. Thus, in the presence of a localized bronchocele of an unknown nature, despite the apparent lack of dynamics according to X-ray data, control studies by contrast-enhanced computed tomography of the chest cavity organs should be conducted at regular intervals to assess reliably the dynamics of changes and exclude bronchial obstruction by a neoplasm.

Computed tomography is a preferred diagnostic method; however, given the peculiarities of the location of typical carcinoids, many authors have recommended fibrobronchoscopy with transbronchial biopsy as complementary imaging methods [4, 5, 15].

Surgical resection is the gold standard for the treatment of typical carcinoids, as this pathology has a low sensitivity to chemotherapy and radiation therapy. In the case of complete endobronchial location of the carcinoid in the central regions, resection can be performed using the transbronchial approach [6, 8, 13].

CONCLUSION

Bronchocele is a benign finding in most cases, but in localized bronchocele, the oncological nature of bronchial obstruction should be ruled out. For this purpose, computed tomography of the chest cavity organs with intravenous contrast enhancement and fibrobronchoscopy with biopsy are recommended.

Some types of neoplasms, such as a typical carcinoid, are characterized by extremely slow growth. Even with negative results on the initial examination of a local bronchocele, these changes require oncological alertness and periodic examinations over time.

ADDITIONAL INFORMATION

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Consent for publication. Written consent was obtained from the patient for publication of relevant medical information and all of accompanying images within the manuscript.

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AUTHORS' INFO

*** Pavel V. Gavrilov,** MD, Cand. Sci. (Med.);
address: 2-4, Ligovskiy pr., Saint-Petersburg, 191036, Russia;
e-mail: spbniifrentgen@mail.ru;
ORCID: https://orcid.com/0000-0003-3251-4084

Kseniya V. Prusakova, MD;
e-mail: ksenya.rush@mail.ru;
ORCID: https://orcid.com/0000-0002-3934-6290

ОБ АВТОРАХ

*** Гаврилов Павел Владимирович,** к.м.н.;
адрес: Россия, 191036, Санкт-Петербург, Лиговский пр., д. 2-4;
e-mail: spbniifrentgen@mail.ru;
ORCID: https://orcid.com/0000-0003-3251-4084

Прусакова Ксения Владимировна;
e-mail: ksenya.rush@mail.ru;
ORCID: https://orcid.com/0000-0002-3934-6290

* Corresponding author / Автор, ответственный за переписку