

Pulmonary sequestration associated with significant elevation of serum carbohydrate antigen 19-9: report of two cases

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Abstract: This report presents two young women were admitted to the department of thoracic surgery, Beijing Chao-Yang Hospital because of the extremely elevation of the serum Carbohydrate antigen 19-9 (CA19-9). The enhanced Computed Tomography scan showed that the lesion of the lung and the anomalous artery, and diagnosis of the pulmonary sequestration was made. The positron emission tomography/computed tomography (PET/CT) scan was taken to exclude any kind of malignant tumor. Both of them had the lobectomy operation confirmed our preoperative diagnosis. After the operation, the serum level of CA19-9 decreased to normal range rapidly. Therefore, in these two cases, the markedly elevation of serum CA19-9 level may have resulted from the pulmonary sequestration.

Keywords: Pulmonary sequestration; carbohydrate antigen 19-9 (CA19-9); positron emission tomography/computed tomography (PET/CT)

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Introduction

Pulmonary sequestration is a rare benign disease which diagnosis mainly based on enhanced chest CT. The most common symptom is refractory lung infection. It is a malformation of lung tissue development, to which blood is supplied by the systemic arteries, the aorta is most common.

Carbohydrate antigen 19-9 (CA19-9) is a widely accepted tumor marker for digestive system tumor. Also, CA19-9 can be elevated slightly in the patients with some certain benign diseases, such as chronic hepatitis, chronic pancreatitis, cholelithiasis, and chronic glomerulonephritis. However the elevation of CA19-9 is rare in benign lung diseases.

In these two cases, the positron emission tomography/computed tomography (PET/CT) was a very effective and necessary method to help making the diagnosis and excluding the malignant tumor before surgery.

Both of the two patients known about the whole process of the diagnosis and treatment, signed the informed consent before surgery and agreed with the postoperative follow-up.

Case presentation

Case 1

A 26-year-old woman was admitted to Beijing Chao-Yang Hospital because of the elevated serum level of the tumor marker CA19-9 without any symptoms. Physical examination showed no abnormalities. Past history: the patient got pneumonia once a year since 9 and treated with antibiotic. The laboratory examination results were normal except the markedly elevation of the CA19-9 (2,095.79 U/mL) at admission, we checked the serum CA19-9 again, the level was still high (2,682.87 U/mL). The enhanced chest and abdomen CT showed the consolidation of S10 segment in right lower lobe and an anomalous artery arising from the descending thoracic aorta (*Figure 1*), without finding any other evidence of digestive system malignant tumor. The patient also took a PET/CT exam to exclude gynecological disease, the result showed that the right lower lobe consolidate with the metabolic increased (SUVmax 3.9), suspected it as a benign lesion, the pulmonary sequestration



Figure 1 Contrast enhanced chest CT, PET/CT and histological result. (A) The S10 segment in right lower lobe consolidation and an anomalous artery arising from the descending thoracic aorta extending into the sequestered lung; (B) the lesion with increased metabolism; (C) histopathology (magnification, $\times 40$) of the resected lung, partial bronchiectasis, interstitial lymphoid follicles formation, no tumor sign. PET/CT, positron emission tomography/computed tomography.

Table 1 The value of the serum level of tumor marker CA19-9

| Tumor marker | Pre-operation | 5 days after operation | 2 months after operation | 6 months after operation |
|---------------|---------------|------------------------|--------------------------|--------------------------|
| CA19-9 (U/mL) | 2,682.87 | 1,098.33 | 68.63 | 12.74 |

CA19-9, carbohydrate antigen 19-9.

was most possible, no other specific abnormality was found. So, the right lower lobe pulmonary sequestration diagnosis was made and a lobectomy operation was performed. The pathology result was not consistent with malignant tumor. After the surgery, the serum CA19-9 level decreased to 68.63 U/mL in 2 months after surgery and to normal level (12.74 U/mL) in 6 months (*Table 1*).

Case 2

A 34-year-old woman came to our clinic with cough and fever for over 1 month, the highest temperature was 38.2 °C. She visited the previous hospital, took the X-ray chest radiograph, the result showed that left lower lobe pneumonia. Laboratory tests results: WBC $15.29 \times 10^9/L$, CRP >200 mg/L, and CA19-9 1,671.3 U/mL. She took a period antibiotic therapy and the temperature dropped to normal while cough aggravated with a lot of purulent sputum. An enhanced chest CT scan indicated the S10 segment in left lower lobe consolidation and an anomalous artery arising from the descending thoracic aorta (*Figure 2*). It was correspond with the typical imaging of the

pulmonary sequestration. Therefore we thought it was benign lesion more likely. The PET/CT scan result confirmed our diagnosis: the left lower lobe irregular consolidation with uneven high metabolic (SUVmax 10.8), suspected the inflammation, but the malignant lesion cannot be excluded. Better to review it after anti-infection treatment. Then the patient came our clinic for the surgery as pulmonary sequestration. We also took some blood test after admission, the serum level of the tumor makers still high: CA19-9 1,414.08 U/mL, CA125 99.67 U/mL, carcino embryonic antigen (CEA) 10.61 ng/mL. A left lower lobe resection was performed. The first day after the operation, we checked the tumor makers again, all the abnormal items decreased sharply: CA19-9 387.97 U/mL, CA125 40.74 U/mL, CEA 2.6 ng/mL. The third day after the operation: CA19-9 252.93 U/mL, CA125 40.12 U/mL, CEA 2.79 ng/mL. The tumor maker decreased to normal in 2 months after the surgery. The pathology showed that the left lower lobe fungal infection with abscess formation (*Table 2*).

Discussion

CA19-9 is a tumor marker which is not so specific for malignant tumor, which can be found in the normal epithelial lining of the biliary tract, gastric mucosa and pancreatic duct (1), or even respiratory epithelium (2). Some reports showed that the CA19-9 is also produced by the bronchiolar epithelia, as a result of airway damage; it can be elevated in serum (3). Bronchial mucus contains large amounts of CA19-9, which appears to be produced in the columnar epithelia of respiratory glands, even if

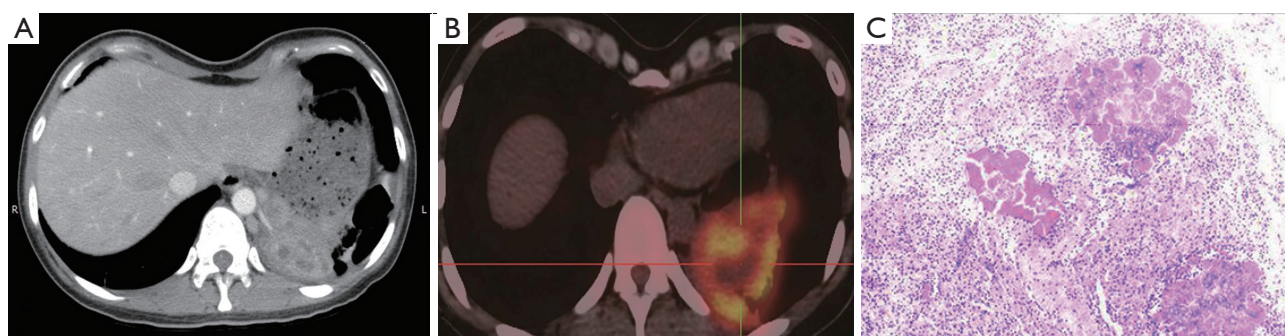


Figure 2 Contrast enhanced chest CT, PET/CT and histological result. (A) The consolidation of S10 segment in left lower lobe and an anomalous artery extending to the lesion from the descending thoracic aorta; (B) hypermetabolism of the sequestered lung; (C) histopathology (magnification, $\times 40$) of the resected lung, no tumor sign as well. Special staining showed that the fungal infection with abscess formation. PAS(+); PASM stain(+). PET/CT, positron emission tomography/computed tomography.

Table 2 The value of the serum level of tumor marker CA19-9, CA125 and CEA

| Tumor marker | Pre-operation | 1 day after operation | 5 days after operation | 15 days after operation | 2 months after operation |
|---------------|---------------|-----------------------|------------------------|-------------------------|--------------------------|
| CA19-9 (U/mL) | 1,414.08 | 387.97 | 252.93 | 90.34 | 20.91 |
| CA125 (U/mL) | 99.67 | 40.74 | 40.12 | 74.08 | 16.46 |
| CEA (ng/mL) | 10.61 | 2.93 | 2.79 | - | - |

CA19-9, carbohydrate antigen 19-9; CEA, carcino embryonie antigen.

the serum CA19-9 level are normal (4). CA19-9 may be synthesized and secreted by normal bronchial epithelial cells, and gradually accumulates in the sequestered lung with no congestion in the normal bronchial tree. The relevant articles have proved the opinion above. Therefore, to avoid the potential misdiagnose, it is important to be aware of the benign lung diseases, especially the pulmonary sequestration associated with the elevated tumor marker level of the serum. Some articles are consistent with the observed association between pulmonary sequestration and the elevated level of serum CA19-9 in this case report. The exact mechanism of the condition remains a controversial subject. Both of the patients are young women, and there are maybe some other factors related with this phenomenon, such as gender, age, position of the lesion, and the level of hormone. Further study on the relationship between the tumor marker and pulmonary sequestration is still required.

This case report demonstrated that the elevation of the serum CA19-9 level may have relationship with the pulmonary sequestration. Examinations, including the PET/CT, detected no other diseases except the pulmonary

sequestration. After the lobectomy operation, the serum CA19-9 level decreased to normal range rapidly. Therefore, it was obvious that the elevated serum CA19-9 level were caused by the pulmonary sequestration. Other studies have also reported similar conclusion. We retrieved 16 English articles from the PubMed for relevant key-word as “pulmonary sequestration & CA19-9” in recent 27 years, of which 14 were reported the elevated CA19-9 in the pulmonary sequestration. Most of the patients took the surgery treatment, and the CA19-9 decreased rapidly after the surgery, and one case did not mention the treatment. But none of them have taken the PET/CT scan exam.

PET/CT is a new imaging technology which combines both of the advantages of the PET (functional metabolic imaging) and CT (anatomical structure imaging) together. It is very useful in discriminating malignant tumor from benign diseases which can show us the metabolism condition of the lesion. In our study, both of the two patients took the PET/CT exam, and the result indicated the pulmonary sequestration instead of the malignant tumor. So we do think the PET/CT scan can systemic

screen and exclude the malignant disease. In our opinion, a PET/CT exam preoperatively will be definitely helpful to diagnose the pulmonary sequestration and also avoid misdiagnosis.

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None.

Footnote

Conflicts of Interest: The authors have no conflicts of interest to declare.

Informed Consent: Written informed consent was obtained from the patient for publication of this manuscript and any accompanying images.

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