

Pulmonary sequestration associated with a synchronous elevation of carbohydrate antigen 50 and 19-9: a case report

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Abstract: This report describes a 37-year-old woman who experienced elevated serum carbohydrate antigen 19-9 (CA19-9) and carbohydrate antigen 50 (CA50). Intralobar pulmonary sequestration was confirmed via enhanced chest computed tomography (CT) scanning and positron emission tomography/computed tomography (PET/CT), which indicated two abnormal vessels arising from the descending thoracic aorta. Lobectomy of the left inferior lobe was performed as the optimal surgical approach and the pathological analysis met the diagnosis of intralobar pulmonary sequestration. Review of the patient's serum levels of CA50 and CA19-9 showed that these two tumor markers significantly decreased after surgery and finally went down to normal values. Therefore, the synchronically significant elevation of serum CA50 and CA19-9 was due to intralobar pulmonary sequestration.

Keywords: Pulmonary sequestration; tumor marker

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Introduction

Carbohydrate antigen 19-9 (CA19-9) and carbohydrate antigen 50 (CA50) are widely considered as a gastrointestinal tumor marker. However, they can also elevate in some other diseases, such as chronic hepatitis, chronic pancreatitis, cholelithiasis, chronic glomerulonephritis, and myelodysplastic syndromes (1).

Pulmonary sequestration is a benign pulmonary disease. Previous researches show that pulmonary sequestration is associated with some tumor markers at times (2), mostly CA19-9 (3). In this case, we are the first to report a synchronically significant elevation of serum CA50 and CA19-9 in pulmonary sequestration.

Case presentation

A 37-year-old woman complained for a significant elevation of CA50 and CA19-9, which was found in an annual medical examination. Computed tomography (CT) scanning of the whole abdomen was performed and

didn't show any positive findings. The patient did not manifest any symptoms, such as cough, hemoptysis and so on. The laboratory examinations revealed a significant elevation of CA50 and CA19-9, which highly reached to 50.02 and 729.48 U/mL respectively. The enhanced CT scanning of the chest (*Figure 1A*) and the CT angiography of the thoracic aorta (*Figure 1B*) showed two abnormal vessels arising from the descending thoracic aorta and consolidation of the posterior basal segment (S10) in the left inferior lobe. To exclude systemic malignant diseases, the positron emission tomography/computed tomography (PET/CT) was performed and didn't show any evidences of malignant signs. The maximum of standardized uptake value (SUVmax) of the consolidation of S10 segment was 4.8 (*Figure 1C*), which was prone to benign lesion. Collectively, pulmonary sequestration was considered as the primary diagnosis. Therefore, we performed a lobectomy of the left inferior lobe through a standard thoracotomy under general anesthesia with a single-lumen endotracheal tube for the patient. The pathological examination confirmed the diagnosis of intralobar pulmonary sequestration (*Figure 1D*).

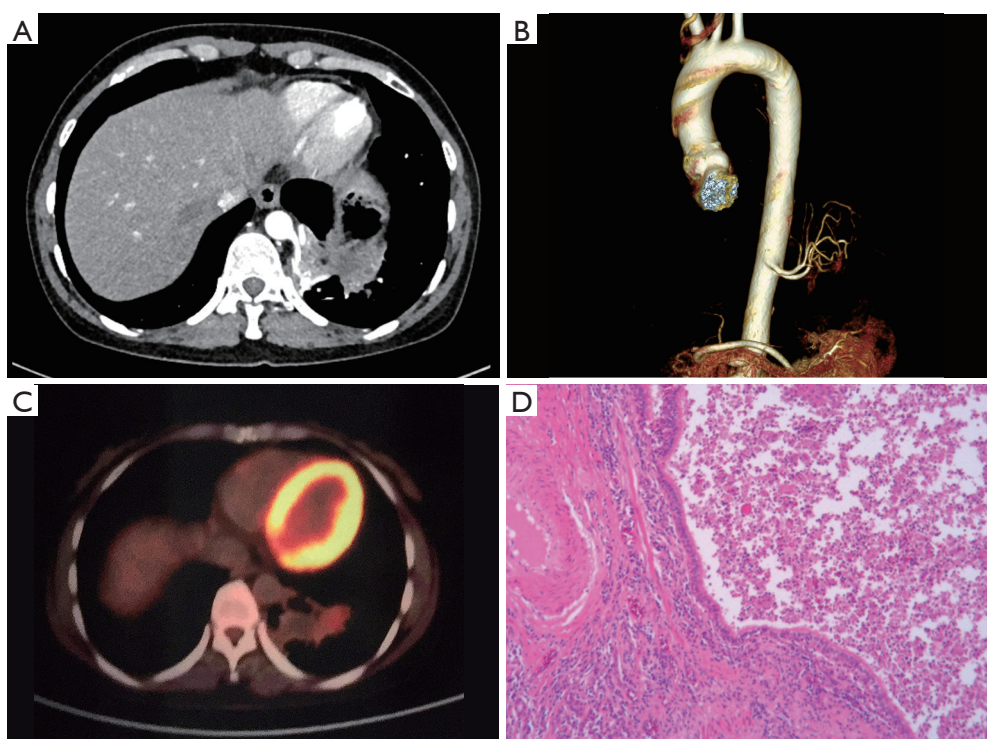


Figure 1 The enhanced CT scanning of the chest, CT angiography of the thoracic aorta, PET/CT and pathological result. (A,B) Enhanced CT scanning of the chest and CT angiography showed two anomalous vessels arising from the descending thoracic aorta; (C) PET/CT revealed slightly enhanced metabolism of the S10; (D) histopathology (magnification, $\times 400$) of the resected lung with interstitial lymphoid follicles formation. CT, computed tomography; PET/CT, positron emission tomography/computed tomography; S10, posterior basal segment.

Table 1 Variation of the serum level of pre-surgery and post-surgery CA 50 and CA19-9

| Tumor marker | Pre-surgery | 5 days after surgery | 1 month after surgery | 3 months after surgery | Reference range |
|---------------|-------------|----------------------|-----------------------|------------------------|-----------------|
| CA50 (U/mL) | 50.02 | 33.07 | 5.75 | 3.59 | 0–25 |
| CA19-9 (U/mL) | 729.48 | 348.78 | 66.21 | 21.1 | 0–37 |

CA 50, carbohydrate antigen 50; CA19-9, carbohydrate antigen 19-9.

Review of serum CA50 and CA19-9 levels showed that these two tumor markers decreased to 33.07 and 348.78 U/mL 5 days after surgery. As expected, they continuously decreased to 5.75 and 66.21 U/mL 1 month after surgery and returned to normal range 3 months after surgery (*Table 1*).

Discussion

CA50 and CA19-9, which are detected by monoclonal antibodies, are non-specific carbohydrate tumor markers. CA50 is commonly associated with gastrointestinal tumors,

such as pancreatic cancer, colorectal cancer, gastric cancer and biliary tract cancer. Moreover, it has been reported that the elevation of CA50 could be found in other carcinomas besides of gastrointestinal tumors, such as non-small cell lung cancer, papillary thyroid carcinoma and myelodysplastic syndrome (4-6). CA19-9 mostly presents in gastrointestinal tumor, such as pancreatic cancer (7) and cholangiocarcinoma (8). Besides, some articles indicate that the elevation of CA19-9 can also be observed in benign diseases, such as uterine fibroids (9) and hydronephrosis (10).

As to pulmonary sequestration, it is a rare benign disease with symptoms of cough and expectoration. Sometimes,

the patient is asymptomatic with complaining for an elevation of tumor markers by screening of the medical examination. The accurate mechanism of the elevated serum tumor markers in pulmonary sequestration is still uncertain. However, some scholars have clarified that CA19-9 could be produced by bronchiolar epithelia after injury of the respiratory tract (11). The bronchial epithelial may synthesize and secrete CA19-9, which stores in the sequestered lung and results in the elevated tumor markers in serum (12).

This case report indicated that the elevation of serum CA50 and CA19-9 may be correlated with pulmonary sequestration. In order to exclude malignant diseases, we performed PET/CT, which showed a negative result of malignant signs. The chest CT scanning showed two anomalous arteries, which gave a primary diagnosis of intralobar pulmonary sequestration. After a surgery of lobectomy, the serum CA50 and CA19-9 level decreased significantly. Three months after surgery, the serum CA50 and CA19-9 level went down to normal range. These results highly indicated the potential relationship between pulmonary sequestration and the abnormal rise of serum CA50 and CA19-9.

To the best knowledge of us, we are the first to report a synchronically significant elevation of serum CA50 and CA19-9 in pulmonary sequestration. Therefore, in case we find any unidentified elevation of the serum CA50 or CA19-9 in asymptomatic patients, excluding malignant diseases, pulmonary sequestration should be considered as a differential diagnosis in our opinions.

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None.

Footnote

Conflict 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.

References

- Ye X, Fu Y, Hu B, et al. Pulmonary sequestration associated with significant elevation of serum carbohydrate antigen 19-9: report of two cases. *Ann Transl Med* 2016;4:373.
- Dong J, Cai Y, Chen R, et al. A Case Report and a Short Literature Review of Pulmonary Sequestration Showing Elevated Serum Levels of Carbohydrate Antigen 19-9. *J Nippon Med Sch* 2015;82:211-5.
- Fontana D, Della Beffa V, Fusca M, et al. Extralobar pulmonary sequestration showing high CA 19-19 levels. *Minerva Chir* 2007;62:39-42.
- Yan GY, Xu ZY, Deng HB, et al. Effects of chemotherapy combined with Chinese herbal medicine Kangliu Zengxiao decoction on tumor markers of patients with advanced non-small-cell lung cancer: a randomized, controlled trial. *Zhong Xi Yi Jie He Xue Bao* 2011;9:525-30.
- Nikiel B, Chekan M, Jaworska M, et al. Expression of the selected adhesive molecules (cadherin E, CD44, LGAL3 and CA50) in papillary thyroid carcinoma. *Endokrynol Pol* 2006;57:326-35.
- Dalamaga M, Karmaniolas K, Kontos F, et al. Clinical study on the serum carcinoembryonic antigen, CA 19-9, CA 50 and alpha-fetoprotein levels in patients with myelodysplastic syndromes. *Leuk Lymphoma* 2006;47:1782-7.
- Lei XF, Jia SZ, Ye J, et al. Application values of detection of serum CA199, CA242 and CA50 in the diagnosis of pancreatic cancer. *J Biol Regul Homeost Agents* 2017;31:383-8.
- Yoo T, Park SJ, Han SS, et al. Postoperative CA19-9 Change Is a Useful Predictor of Intrahepatic Cholangiocarcinoma Survival following Liver Resection. *Dis Markers* 2015;2015:298985.
- Ma Y, Shao X. Uterine fibroids with positive 18F-FDG PET/CT image and significantly increased CA19-9: A case report. *Medicine (Baltimore)* 2017;96:e9421.
- Filipovic B, Milinić N, Gacic J, et al. Benign Hydronephrosis and Elevated of Serum Levels of Carbohydrate Antigen CA 19-9: A Case Report. *Am J Case Rep* 2016;17:395-7.
- Ambiru S, Nakamura S, Fukasawa M, et al. Intralobar pulmonary sequestration associated with marked elevation of serum carbohydrate antigen 19-9. *Ann Thorac Surg* 2009;88:2010-1.
- Komatsu H, Mizuguchi S, Izumi N, et al. Pulmonary sequestration presenting elevated CA19-9 and CA125 with ovarian cysts. *Ann Thorac Cardiovasc Surg* 2014;20 Suppl:686-8.

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