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Case report

Primary salivary gland tumors of the lung: Two cases date report and literature review

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ABSTRACT

Background: Primary salivary gland-type tumors of the lung (PSGTTL) is a rare intrathoracic malignant tumor that accounts for all lungs <1% of tumors.
Purpose: To introduce two case reports of primary lung salivary gland tumors, and highlight their diagnosis and treatment challenges.
Case report: The first case was a 30-year-old female, who complained of repeated coughing and dyspnea for 1 year and worsening for 2 days. Chest CT and bronchoscopy showed new organisms in the lower part of the trachea, that the bronchus obstruction accounted for 70%. The biopsy histology revealed a adenoid cystic carcinoma. She underwent extensive surgical resection and multiple radiotherapy, and She is recovering well from follow-up. The second case was a 70-year-old man, who complained of intermittent sputum blood for 2 years, worsening hemoptysis and chest tightness for 3 months. The new organisms was found in the upper trachea from Chest CT and bronchoscopy, and histological biopsy was used to diagnose epithelial myoepithelial carcinoma. He underwent twice bronchoscopy thermal ablation treatments. The follow-up is currently in good condition.
Conclusion: Primary lung salivary gland tumors are considered to be rare malignant tumors in the lungs. Early detection is advocated as late presentation with advanced tumor presents diagnostic and therapeutic challenges.

1. Preface

Primary salivary gland-type tumors of the lung (PSGTTL) are rare within trathoracic malignancies, accounting for <1% of all lung tumors [1]. In the 2015 WHO classification of lung tumors, this type of tumor includes adenoid cystic carcinoma (ACC), mucoepidermoid carcinoma (MEC), myoepithelioma carcinoma (EMC) and Pleomorphic adenoma, which are four major types [2]. At present, there is no uniform standard for PSGTTL treatment. Some studies have pointed out that surgical resection is the best initial treatment option. However, bronchoscopy intervention may be used as initial treatment to patients who are clinically at a higher clinical stage, or as an initial treatment transition therapy before surgery [3].

2. Case 1

The patient, who is a 29-years-old female and a farmer, has no history of chronic disease or smoking. She has no history of tuberculosis and contact with tuberculosis, no personal or family tumor history. Simultaneously, The individual has no history of surgery. She was admitted to the Fourth Affiliated Hospital of Anhui Medical University on September 14, 2019. She complained of repeated coughing and dyspnea for 1 year and worsening for 2 days. On September 13, 2019, a chest CT examination of a local hospital found a space occupying image on the upper carina. After admission to our hospital, enhanced chest CT + tracheal reconstruction suggested new organisms on the upper carina (Fig. 1-A, 1-B). On September 18, 2019, She experienced a bronchoscopy, which revealed a grape cluster-like new creature on the carina at 3 o'clock on the right wall of the lower trachea. The opening of the right main bronchus was covered by the new creature, and the 1-TQ290 bronchoscope of Olympus could not pass (Fig. 1-D). At the same time, There were many purulent secretions in left bronchus. After aspiration, the left mucosa was smooth, the left lumen was unobstructed, and no new organisms were seen. A new biological biopsy will be sent for histopathological examination. Immunohistochemistry showed: CK (+), SMA (+), Calponin (partial +), CD117 (+), P63 (+), S-100 (focal +), TTF-1 (−); in line with adenoid Adenoid cystic carcinoma cancer (ACC) (Fig. 1-E1-F.1-G). We give her a preliminary diagnosis of primary

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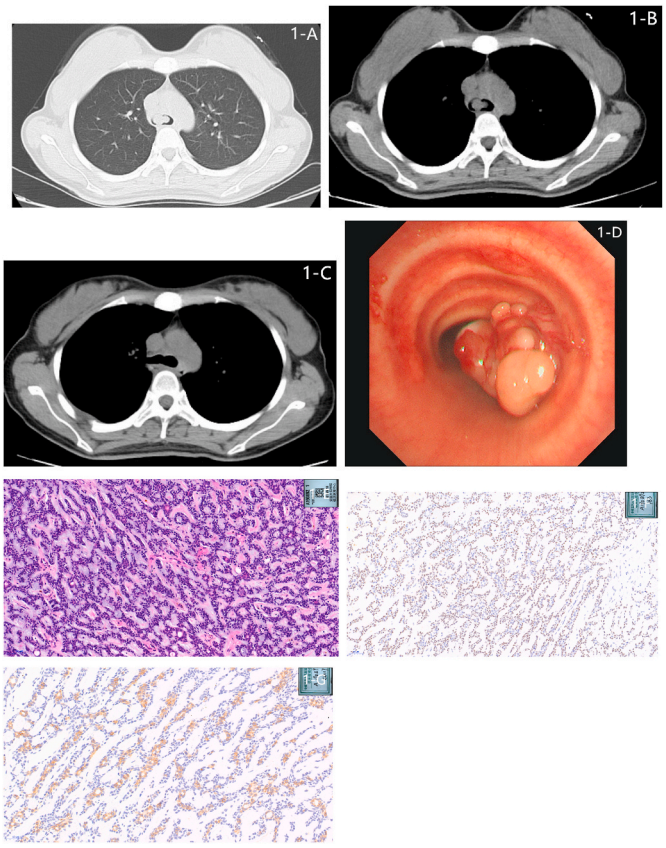

Fig. 1. Imaging of case 1.

salivary gland lung tumor (PSGTTL). We organized a Multidisciplinary consultation, including the oncology, the pathology, the thoracic surgery, and the radiology departments are invited. After discussion, We Consider that the patient was relatively young and has obvious symptoms of airway obstruction, and Imaging shows that the local tracheal cartilage ring was destroyed in the lesion. Simple interventional treatment under bronchoscope was likely to cause tracheal fistula. Surgery was recommended. Subsequently, the patient had a surgical operation in Shanghai Thoracic Hospital, which removed her section of the diseased trachea. The Histopathological examination of that showed: the tumor size was 2.8cm*2.5cm*2.3cm, the tracheal wall was fully infiltrated, and the adventitia was involved, including nerve invasion. "Left common bronchus cut end" and "right common bronchus cut end" have no cancer involvement; P63 (part +), CK5/6 (part +), CK (epithelial +), CD117 (part +), Calponin (part +), CK7 (partial +), KI-67 (10% +), consistent with adenoid cystadenocarcinoma.And after multiple radiotherapy treatments, the patient had no obvious respiratory symptoms and had no obvious new biological manifestations in the trachea on the chest CT on November 2, 2019 (Fig. 1-C). Follow-up will continue.

1-A. 1-B: New biological shadows in the lumen above the tracheal carina; 1-C. After the operation, the lumen of the carina is unobstructed and no new biological shadows are seen.; 1-D Bronchoscopy findings: New cauliflower-like organisms in the trachea highly block the lumen 0.1-E: HE staining of histopathology showed that the tumors were distributed in a sieve shape, and the cell nests were cylindrical microcapsule spaces, which contained hyalin or basophilic mucus-like substances; 1-F: Immunohistochemistry of P63(+); 1-G: Immunohistochemistry of CD117 (+).

3. Case 2

The other patient, who is a 70-years-old male and a farmer too. He had a history of hypertension and a history of smoking 1000 cigarettes per year. He had no history of tuberculosis, no contact with tuberculosis. No personal or family history of cancer and No history of surgery. On September 5, 2020, He lived in the Fourth Affiliated Hospital of Anhui Medical University. The chief complaint of intermittent sputum and blood for more than 2 years, worsened by 3 months. His sputum and blood were intermittent, as early as 2018, he had a hemoptysis, accompanied by coughing up a little carrion tissue sample, which was not taken seriously at that time. This time, phlegm and blood recurred and worsened in the past 3 months, accompanied by coughing up carrion tissue samples again. The course of the illness was accompanied by chest tightness after exercise, throat wheezing, no fever, night sweats, and no significant weight loss. On physical examination, his temperature was normal, his breathing rate was normal, and his lungs were auscultated without rales and wheezing. One week ago, the chest CT examination of the external hospital on 2020.8.22 found that the anterior wall of the upper trachea was tumor with calcification. Bronchoscopy revealed new organisms in the upper trachea. The preliminary pathological report of biopsy indicated: polymorphic adenoma. He was admitted to our hospital for further diagnosis and treatment. After admission to our hospital, the three-dimensional reconstruction of the trachea was perfected. Tips: the anterior wall mass of the upper trachea is accompanied by local calcification (Fig. 2-A,2-B). We discussed the treatment options, and finally recommended that the patient undergo bronchoscopy for airway examination, re-biopsy of endobronchial lesions and possible therapeutic interventions.

Bronchoscopy showed that the anterior wall of the main trachea was 5cm from the glottis and the cauliflower-like neoplasms partially blocked the lumen and the base was wide(Fig. 2-E). The mass in the trachea is very large, in the bronchus, the distal left and right bronchial mucosa are smooth, the lumen is unobstructed, and no new organisms are seen. The lesions are pretreated with epinephrine and cold saline. Then use an electrocautery snare, biopsy forceps and aspirator to remove the tumor piece by piece. Use argon plasma coagulation (APC) to stop bleeding for tumor coagulation. The excised tissue is submitted for histopathological examination. Microscopic observation (Fig. 2-G2-H.2-I) showed that the immune group: tumor cells CK7 (glandular epithelium +), CD117 (glandular epithelium +), CK8 (glandular epithelium +), TTF (−), S-100 (partial +), CK5/6 (myoepithelial+), SMA (myoepithelial+), P63 (myoepithelial+), P40 (myoepithelial+), syn (−), CgA (−), CD56 (−), ki-67 (+, About 10%). magnetic resonance imaging (MRI) Examination of the head revealed no salivary gland pathologies.The discovery can diagnose primary epithelial myoepithelial carcinoma (EMC) of the lung. After 5 days, the bronchoscopy showed that the tumor was significantly smaller than before, and the trachea was smoother than before, and the necrosis on the tumor surface was cleaned up(Fig. 2-F). After bronchoscopy heat treatment, His chest tightness and cough symptoms were significantly reduced, and he was discharged from the hospital. Outpatient follow-up was planned, and if necessary, bronchoscopy and treatment under the microscope again.

2-A.2-B. Neotracheal imaging with calcification; 2-C.2-D: After bronchoscopy intervention, the intratracheal neoplasia is smaller than before, and the lumen is smoother than before; 2-E: Before treatment of the bronchoscope, new organisms in the trachea partially block the lumen; 2-F:After bronchoscope thermal treatment, the new organisms shrink and the trachea is smoother than before; 2-G: HE staining of histopathology shows double-layer duct-like tissue, the inner layer is lined with cubic cells and the cytoplasm is small; the outer layer is composed of myoepithelial cells with clear cytoplasm; 2-H: Immunohistochemistry of P63 (+); 2-I: Immunohistochemistry of CD117 (+).

4. Discussion

Due to the basic structural homology between many exocrine glands, salivary gland tumors can occur in many organs. Primary salivary gland-type tumors of the lung (PSGTTL) are rare among intrathoracic

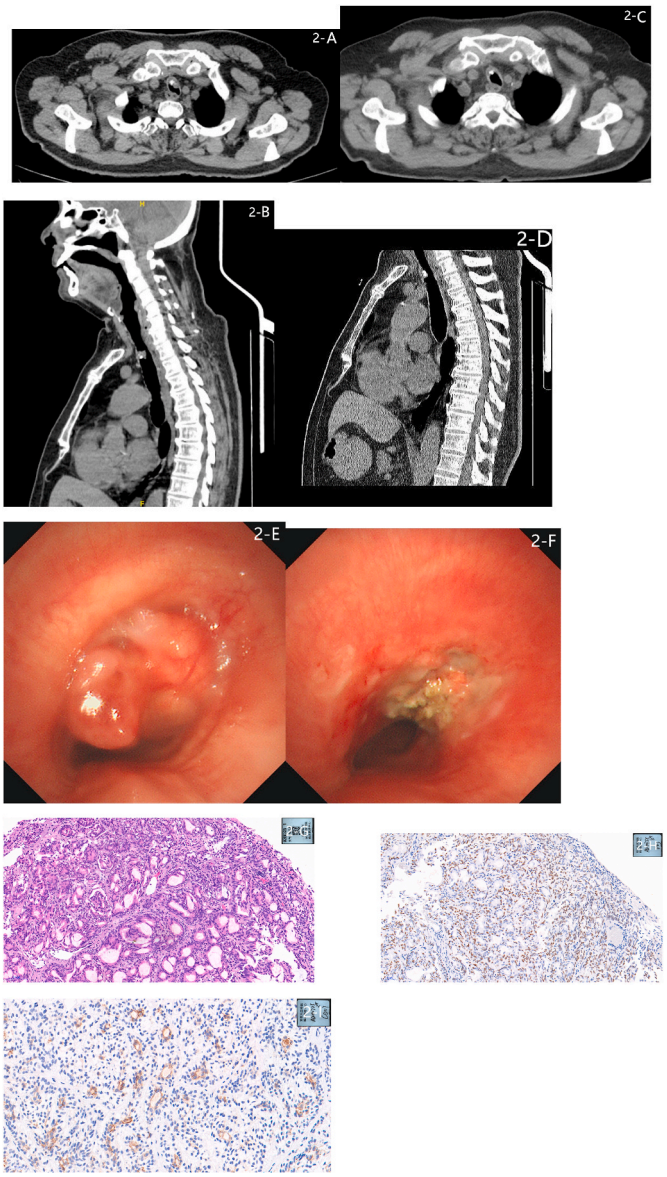

Fig. 2. Imaging of case 2.

malignancies, accounting for <1% of all lung tumors [1]. The differentiation of PSGTTL and salivary gland tumors with lung metastasis mainly depends on the clinic, whether there is a history of salivary gland tumors, and whether the salivary gland tumors are found in imaging examination. PSGTTL has no different from its saliva-derived counterpart in terms of histologically. So It is maybe that PSGTTLcomes from the submucosal gland of the tracheobronchial tree [4]. In the 2015 WHO classification of lung tumors, this type of tumor includes adenoid cystic carcinoma (ACC), mucoepidermoid carcinoma (MEC), myoepithelioma carcinoma (EMC) and Pleomorphic adenoma, which are four major types [2].The study found that in addition to the pleomorphic adenoma, MEC is the most common pathological type among the other three types PSGTTL, followed by ACC [5,6]. According to its statistics, in PSGTTL, the proportion of tumors located in the central airway is 15.9%–100%. The clinical manifestations depend to a large extent on the location of the tumor and the condition of the distal obstruction. Most patients have cough and difficulty breathing; The clinical symptoms of others are hemoptysis, wheezing or obstructive pneumonia. PSGTTL is mainly of the central type. Therefore, it has long been believed that the tracheal glands of the trachea and bronchial submucosa are the origin of this type of tumor. However, there are reports in the literature that the case is located under the visceral membrane [7–9]. Therefore, some scholars speculate that this type of tumor originated from lung primitive stem cells.

Pandey et al. [10]. reported that most patients were initially misdiagnosed as tuberculosis and prescribed anti-tuberculosis drugs, resulting in a delay in the correct diagnosis of PSGTTL. PSGTTL is generally considered to be slow growing and rarely metastasizes when it first appears. Research by Zhu Fen et al. [6]. found that low-grade PSGTTL has better survival time, while high-grade malignant recurrence rate, lung parenchymal invasion and mortality are higher.

PSGTTL and the corresponding primary tumors of the salivary glands, although the two are similar in histology, immunophenotype, genotype, and prognosis, there are also differences. According to reports, most primary adenoid cystic carcinomas of the salivary glands have MYB gene translocations, while primary ACC of the lung have only rare cases where MYB gene translocations have been detected [11]. Patients with primary lung MEC have a good prognosis. For example, Amit.et al. reported that a 26-year-old man, who was cough, sputum and blood, was diagnosed as primary lung MEC by bronchoscopy, and then had a good prognosis after total left lung resection [9]; While the prognosis of patients with primary ACC of the lung is worse than that of patients with primary salivary glands.

In this article, the two cases of PSGTTL are both progressed slowly, located in the central airway, and showed polypoid masses under the bronchoscopy. This type of cancer is rare. Most of the PSGTTL cases reported in the literature are case reports, most of which were removed by surgery [12–15], and a small number of cases were treated by endobronchial intervention.This management strategy is most likely due to its standardized salivary salivary gland tumor counterpart, which usually requires extensive surgical resection. In the two patients in this article, the first case is because the patient is relatively young. It is recommended that the surgical treatment has a good postoperative recovery and a good prognosis to avoid recurrence. In case two, the patient was very old and had a high risk of surgery. Through bronchoscopy intervention, he successfully completed tumor reduction and unobstructed airway. It is worth noting that due to the low incidence of these tumors and the lack of literature, there is no standard of treatment in the treatment of these individuals. Bronchoscopy intervention is used to successfully treat central airway obstruction caused by benign or malignant tumors. Thermal treatments for tumor ablation or coagulation in the airway include electrocautery, argon plasma coagulation (APC), and cryotherapy [16,17].Our two patients initially underwent bronchoscopy for diagnosis and treatment purposes, excised tumors for pathological examination, and confirmed the diagnosis. After the imaging examination has no lymph node metastasis, and the pathological diagnosis is with a known low risk of metastasis, multiple bronchial examinations and treatments can be passed at this time.

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Declaration of competing interest

ZL conceived and designed the structure of the article, HHC collected case data and drafted the manuscript, CXH participated in the bronchoscopy treatment of the two patients, MG and WYJ analyzed the pathological data in the case and provided pictures, WM helped HHC complete the manuscript. All authors read and approve the final manuscript.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.rmcr.2020.101333.

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