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World J Clin Cases 2021 April 26; 9(12): 2696-2950



MINIREVIEWS

- 2696 Standardization of critical care management of non-critically ill patients with COVID-19
Wang CS, Gao Y, Kang K, Fei DS, Meng XL, Liu HT, Luo YP, Yang W, Dai QQ, Gao Y, Zhao MY, Yu KJ
- 2703 Mediastinal lymphadenopathy in COVID-19: A review of literature
Taweasedt PT, Surani S
- 2711 Polycystic ovary syndrome: Pathways and mechanisms for possible increased susceptibility to COVID-19
Ilias I, Goulas S, Zabuliene L

ORIGINAL ARTICLE**Clinical and Translational Research**

- 2721 Circulating tumor cells with epithelial-mesenchymal transition markers as potential biomarkers for the diagnosis of lung cancer
Jiang SS, Mao CG, Feng YG, Jiang B, Tao SL, Tan QY, Deng B

Retrospective Study

- 2731 Management and implementation strategies of pre-screening triage in children during coronavirus disease 2019 pandemic in Guangzhou, China
Shi X, Cai YT, Cai X, Wen XL, Wang JY, Ma WC, Shen J, Wu JX, Liu HY, Sun J, He PQ, Lin Y, Zhao DY, Li PQ
- 2739 Clinicopathological features of superficial CD34-positive fibroblastic tumor
Ding L, Xu WJ, Tao XY, Zhang L, Cai ZG
- 2751 Application of a rapid exchange extension catheter technique in type B2/C nonocclusive coronary intervention *via* a transradial approach
Wang HC, Lu W, Gao ZH, Xie YN, Hao J, Liu JM

SYSTEMATIC REVIEWS

- 2763 Paradoxical relationship between proton pump inhibitors and COVID-19: A systematic review and meta-analysis
Zippi M, Fiorino S, Budriesi R, Micucci M, Corazza I, Pica R, de Biase D, Gallo CG, Hong W

META-ANALYSIS

- 2778 Predictive risk factors for recollapse of cemented vertebrae after percutaneous vertebroplasty: A meta-analysis
Ma YH, Tian ZS, Liu HC, Zhang BY, Zhu YH, Meng CY, Liu XJ, Zhu QS

CASE REPORT

- 2791** Malignant pheochromocytoma with cerebral and skull metastasis: A case report and literature review
Chen JC, Zhuang DZ, Luo C, Chen WQ
- 2801** Unresectable esophageal cancer treated with multiple chemotherapies in combination with chemoradiotherapy: A case report
Yura M, Koyanagi K, Hara A, Hayashi K, Tajima Y, Kaneko Y, Fujisaki H, Hirata A, Takano K, Hongo K, Yo K, Yoneyama K, Tamai Y, Dehari R, Nakagawa M
- 2811** Role of positron emission tomography in primary carcinoma ex pleomorphic adenoma of the bronchus: A case report
Yang CH, Liu NT, Huang TW
- 2816** Positive reverse transcription-polymerase chain reaction assay results in patients recovered from COVID-19: Report of two cases
Huang KX, He C, Yang YL, Huang D, Jiang ZX, Li BG, Liu H
- 2823** Laryngeal myxoma: A case report
Yu TT, Yu H, Cui Y, Liu W, Cui XY, Wang X
- 2830** Prostate stromal tumor with prostatic cysts after transurethral resection of the prostate: A case report
Zhao LW, Sun J, Wang YY, Hua RM, Tai SC, Wang K, Fan Y
- 2838** Intramuscular hematoma in rhabdomyolysis patients treated with low-molecular-weight heparin: Report of two cases
Yuan SY, Xie KF, Yang J
- 2845** Partial response to Chinese patent medicine Kangliu pill for adult glioblastoma: A case report and review of the literature
Sun G, Zhuang W, Lin QT, Wang LM, Zhen YH, Xi SY, Lin XL
- 2854** Behcet's disease manifesting as esophageal variceal bleeding: A case report
Xie WX, Jiang HT, Shi GQ, Yang LN, Wang H
- 2862** Successful endoscopic surgery for emphysematous pyelonephritis in a non-diabetic patient with autosomal dominant polycystic kidney disease: A case report
Jiang Y, Lo R, Lu ZQ, Cheng XB, Xiong L, Luo BF
- 2868** Robotically assisted removal of pelvic splenosis fifty-six years after splenectomy: A case report
Tognarelli A, Faggioni L, Erba AP, Faviana P, Durante J, Manassero F, Selli C
- 2874** Pulmonary alveolar proteinosis complicated with nocardiosis: A case report and review of the literature
Wu XK, Lin Q
- 2884** Detection of EGFR-SEPT14 fusion in cell-free DNA of a patient with advanced gastric cancer: A case report
Kim B, Kim Y, Park I, Cho JY, Lee KA

- 2890** Timing of convalescent plasma therapy-tips from curing a 100-year-old COVID-19 patient using convalescent plasma treatment: A case report
Liu B, Ren KK, Wang N, Xu XP, Wu J
- 2899** Torsades de pointes episode in a woman with high-grade fever and inflammatory activation: A case report
Qiu H, Li HW, Zhang SH, Zhou XG, Li WP
- 2908** Salivary duct carcinoma of the submandibular gland presenting a diagnostic challenge: A case report
Uchihashi T, Kodama S, Sugauchi A, Hiraoka S, Hirose K, Usami Y, Tanaka S, Kogo M
- 2916** Allogeneic hematopoietic stem cell transplantation in a 3-year-old boy with congenital pyruvate kinase deficiency: A case report
Ma ZY, Yang X
- 2923** Congenital bilateral cryptorchidism in an infant conceived after maternal breast cancer treatment: A case report
Hu WK, Liu J, Liu RX, Liu XW, Yin CH
- 2930** Sclerosing polycystic adenosis of the submandibular gland: Two case reports
Wu L, Wang Y, Hu CY, Huang CM
- 2937** Budd-Chiari syndrome associated with liver cirrhosis: A case report
Ye QB, Huang QF, Luo YC, Wen YL, Chen ZK, Wei AL
- 2944** Separated root tip formation associated with a fractured tubercle of dens evaginatus: A case report
Wu ZF, Lu LJ, Zheng HY, Tu Y, Shi Y, Zhou ZH, Fang LX, Fu BP

ABOUT COVER

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Role of positron emission tomography in primary carcinoma ex pleomorphic adenoma of the bronchus: A case report

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Abstract

BACKGROUND

Primary carcinoma ex pleomorphic adenoma arising from the tracheobronchial system is rarely reported.

CASE SUMMARY

We present a patient with primary carcinoma ex pleomorphic adenoma of the bronchus and review the associated literature for further comparison, including age, clinical manifestations, and diagnostic process. This patient had no history of neoplasms of the salivary gland.

CONCLUSION

Positron emission tomography played an important role in the staging work-up of primary carcinoma of ex pleomorphic adenoma. Long-term follow-up was necessary for further prognosis analysis.

Key Words: Primary carcinoma ex pleomorphic adenoma; Positron emission tomography; Bronchus; Lung; Metabolically active region; Case report

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Core Tip: Positron emission tomography plays an important role in defining the size of the malignant component of primary carcinoma ex pleomorphic adenoma and affects

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INTRODUCTION

Carcinoma ex pleomorphic adenoma (Ca ex PA) is a carcinoma arising from a primary or recurrent benign pleomorphic adenoma. Ca ex PA is uncommon, and it has a prevalence rate of 5.6 cases per 100000 malignant neoplasms and a yearly incidence rate of 0.17 tumors per 1 million persons. Cancer predominantly presents in the sixth to eighth decades of life and is slightly more common in females^[1]. Although rarely reported, primary Ca ex PA can arise only from the respiratory tract and manifests respiratory symptoms. Only 11 cases have been reported since 1988^[2].

Due to its rarity, there is no consensus on the diagnostic protocol for Ca ex PA. It is difficult to discuss radiologic features, particularly features of positron emission tomography (PET). The case reported herein only developed hemoptysis without cough or other respiratory symptoms. This patient is the youngest adult among the patients with Ca ex PA reported to date.

CASE PRESENTATION

Chief complaints

Blood-tinged sputum was observed in June 2018, which progressed to bloody sputum 2 mo later.

History of present illness

The 40-year-old married man noticed blood-tinged sputum beginning in June 2018, which progressed to bloody sputum 2 mo later. Therefore, he visited a district hospital where chest computed tomography (CT) showed an irregular mass over the left upper lobe of the lung with suspected pulmonary vessel invasion. In addition, endobronchial lesions caused obstructive pneumonitis and partial atelectasis of the left upper lobe of the lung (Figure 1). Bronchoscopy with cytology and biopsy was performed, and the pathological report demonstrated carcinoid tumors of the lung. Due to the impression of a carcinoid tumor of the bronchus of the left upper lobe of the lung, cT4NxMx, stage IIIA at least, he was referred to our medical center for further staging work-up and pre-operative evaluation.

History of past illness

The patient had no systematic disease or surgical history.

Personal and family history

He was a non-smoker and did not consume alcohol. His family history was unremarkable.

Physical examination

The patient was considered to have a moderate nutritional status with good performance. Body weight was 80.9 kg, body temperature was 36.3 °C; respiration rate was 12 breaths/min, and heart rate was 72 bpm. Neck and head showed no palpable masses or nodules. Back and spine demonstrated no knocking tenderness.

Chest and lungs examination: No chest wall deformity, no accessory muscle respiration, and no surgical scars were observed. Auscultation showed symmetrical and free expansion, and coarse breathing sounds in the left upper field. Percussion revealed no abnormal dullness. Palpation showed symmetrical and free expansion, no

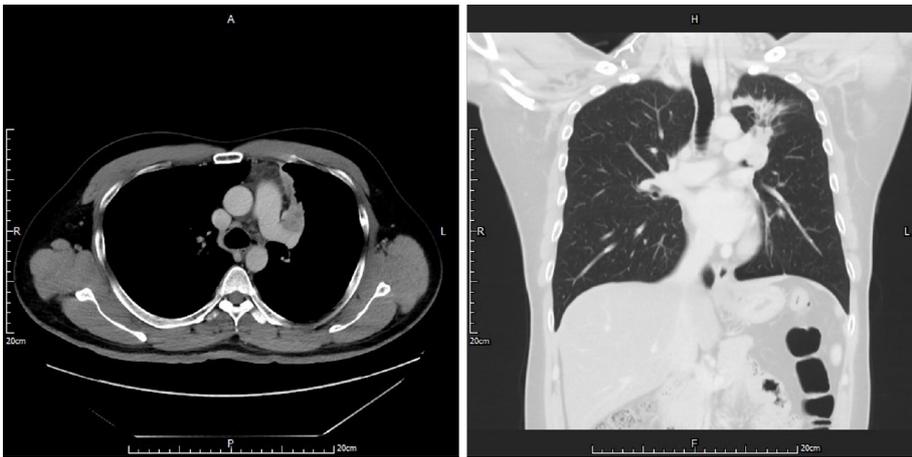


Figure 1 Chest computed tomography showed a tumor located in the left upper lobe of the lung with suspected endobronchial lesions and obstructive pneumonitis. Vessel invasion could not be excluded.

tender points, and no palpable masses. The abdomen revealed normoactive bowel sounds, and was soft without tenderness.

Laboratory examinations

Routine blood tests and blood biochemistry showed no significant abnormalities. Serum levels of tumor markers including carcinoembryonic antigen and squamous-cell-carcinoma antigen were within the normal range.

Imaging examinations

Chest CT showed an irregular mass over the left upper lobe of the lung with suspected pulmonary vessel invasion. In addition, endotracheal lesions caused obstructive pneumonitis and partial atelectasis of the left upper lobe of the lung.

Contrast-enhanced magnetic resonance imaging of the brain showed no evidence of metastasis. Abdominal sonography showed no evidence of intra-abdominal metastasis.

PET showed: (1) A focally intense fluorodeoxyglucose-avid lesion near the left upper lobe bronchus (maximal standard uptake value = 7.1; metabolic size: 2.0 cm) was seen, compatible with lung cancer (cT2 at least, involving the bronchus); and (2) No metastasis/malignancy associated abnormal fluorodeoxyglucose uptake was noted (Figure 2). Bronchoscopy showed an endobronchial tumor.

FINAL DIAGNOSIS

Ca ex PA of the left upper lobe of the lung, pT2aN0M0, stage IB.

TREATMENT

Video-assisted thoracic surgery with lobectomy was performed. One endobronchial, firm and solid tumor measuring 2.2 cm in diameter without a satellite tumor nodule was found in the main bronchus with bronchial cartilage invasion. Due to the clinically close margin, he was treated with a series of concurrent chemo-radiotherapy after surgery.

OUTCOME AND FOLLOW-UP

Follow-up examinations 1 year after surgery showed no disease recurrence or metastasis. Although the serum carcinoembryonic antigen level was within normal limits at initiation, it continued to decline.



Figure 2 Whole-body positron emission tomography scan. A smaller diameter of the metabolically active region of the tumor compared with the tumor size was revealed by chest computed tomography.

DISCUSSION

Primary Ca ex PA of the trachea and bronchi is extremely rare. Only 11 cases have been reported. A comparison with our case was made and the results are as follows:

The average age at operation was 59 years and ranged from 44 years to 71 years. Most of the patients were in their sixties^[2]. Our patient at 40 years old is the youngest adult to date to be diagnosed and surgically managed.

Symptoms might not be important clues for diagnosis. Although dyspnea and cough seem to be common, our case presented with only hemoptysis.

Chest CT showed mild atelectasis and mild obstructive pneumonitis of the left upper lobe of the lung caused by endobronchial partial obstruction. A description of the radiologic manifestations of Ca ex PA is rare. This should be included in the differential diagnosis when an endobronchial lesion with a lung mass is noted.

It is difficult to determine the malignant tumor size from the whole mass; thus initially, the clinical stage was cT4N0M0 as revealed by contrast-enhanced thoracic CT, with probable invasion of the pulmonary arterial branch. However, PET showed only 2 cm of metabolic tumor volume without evidence of lymphadenopathy or distant metastasis. Therefore, surgical intervention was an option. PET plays an important role in both the detection of lymphadenopathy and distant metastasis of lung cancer. In addition, it can measure the true size of the malignant component of this type of tumor. The pathology report showed that the malignant component of the tumor was 2.2 cm in diameter without lymph-vascular invasion, compatible with a T2a lesion.

The malignant component of Ca ex PA demonstrated a wide spectrum of histologic differentiation that needs to be recognized as such in order not to be dismissed as a non-small cell carcinoma or poorly differentiated carcinoma. They usually show myoepithelial differentiation, but not adenocarcinoma or salivary duct carcinoma, as commonly seen in tumors arising in the salivary glands^[3]. Most likely due to the limitation of inappropriate endobronchial specimens taken during bronchoscopy, the gross morphology of the tumor, hemoptysis as the only symptom and its acute onset, and the patient's age, the initial impression was a carcinoid lung tumor. However, the final pathology from lobectomy showed a subepithelial solid tumor composed of obvious epithelial cells forming a duct-like structure (Figure 3A), occasional mucin production, and a focal myoepithelial cell component (reactive to S-100 protein, Figure 3D) with clear cytoplasm, accompanied by chondromyxoid and hyalinizing matrix with bronchial cartilage invasion (Figure 3B). Negative reactivity to chromogranin A and weakly positive reactivity to synaptophysin in tumor cells indicated that a carcinoid lung tumor was unlikely. Therefore, the final diagnosis was Ca ex PA (Figure 3).

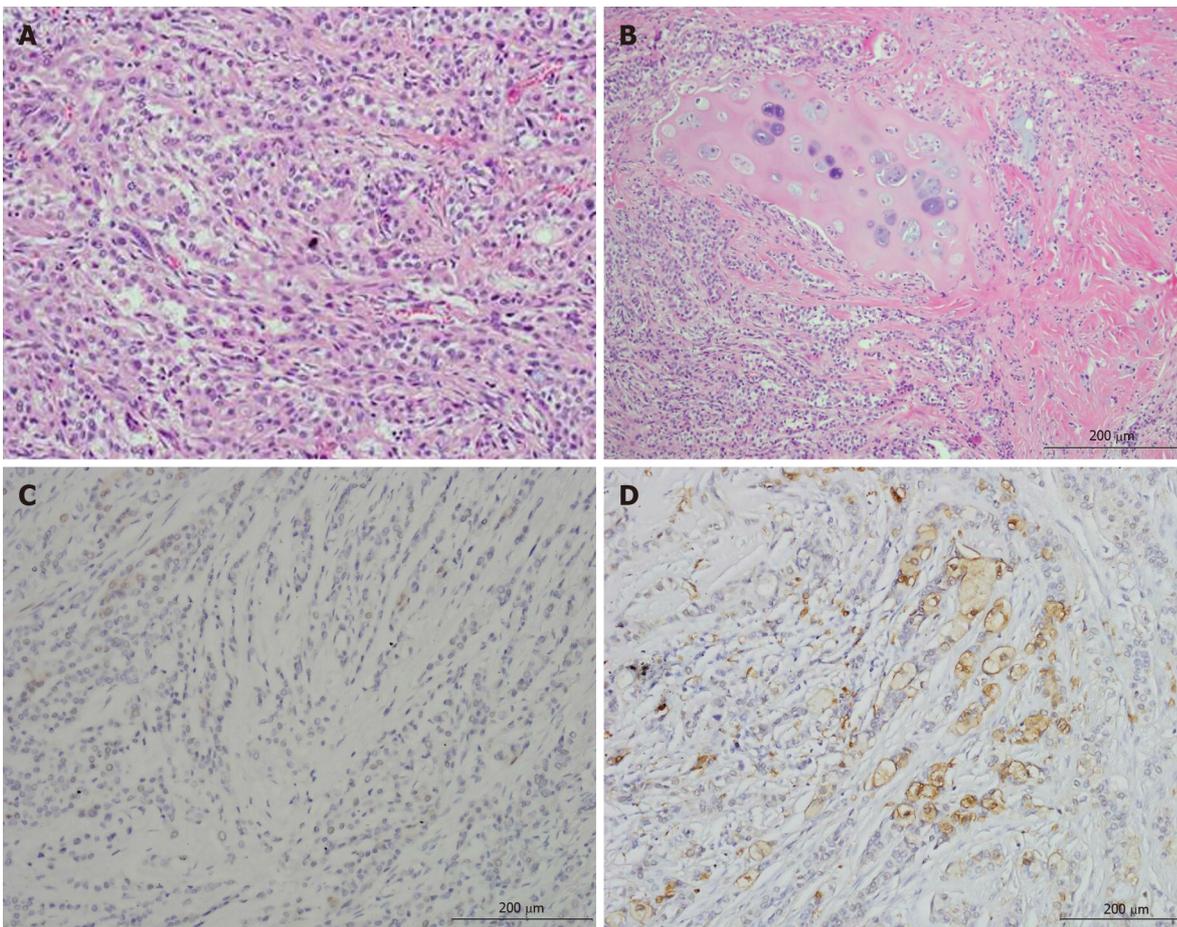


Figure 3 Pathology images. A: Epithelial cells formed a duct-like structure, with occasional mucin production; B: Chondromyxoid and hyalinizing matrix as the background, and bronchial cartilage invasion were found; C: Weakly positive reactivity to synaptophysin in tumor cells indicated that a carcinoid lung tumor was unlikely; D: Myoepithelial cells positive for S-100 protein.

CONCLUSION

Here, we report a case of primary Ca ex PA arising from the main bronchus of the left upper lobe of the lung. Additionally, we show that PET may provide some information on the resectability and true size of the malignant component of this type of tumor. Long-term follow-up for analysis of prognosis is necessary.

REFERENCES

- 1 **Antony J**, Gopalan V, Smith RA, Lam AK. Carcinoma ex pleomorphic adenoma: a comprehensive review of clinical, pathological and molecular data. *Head Neck Pathol* 2012; **6**: 1-9 [PMID: [21744105](#) DOI: [10.1007/s12105-011-0281-z](#)]
- 2 **Xu JJ**, Wang JW, Zhang MM. Primary carcinoma ex pleomorphic adenoma of the bronchus: report of two cases and review of the literature. *Int J Clin Exp Med* 2017; **10**: 3860-3863
- 3 **Weissferdt A**, Moran CA. Pulmonary salivary gland-type tumors with features of malignant mixed tumor (carcinoma ex pleomorphic adenoma): a clinicopathologic study of five cases. *Am J Clin Pathol* 2011; **136**: 793-798 [PMID: [22031319](#) DOI: [10.1309/AJCP50FBZWSACKIP](#)]



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