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Case Report

Mucoepidermoid lung carcinoma in a pediatric patient confused with pneumonia ☆,☆☆

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ABSTRACT

Mucoepidermoid carcinoma (MEC) is a common type of salivary gland malignancy; however, rarely, MEC can arise from the lung. This disease has a non-specific presentation and is often overlooked. Histologically, MEC can be classified into low-grade and high-grade forms. Surgical resection is the optimal treatment for low-grade tumors. In this article, we report a case of MEC in a 5-year-old girl who was initially misdiagnosed with pneumonia. The histological results revealed MEC. Thus, clinicians and radiologists should consider the possibility of this rare entity in patients who fail to respond to antibiotic treatments, even among the pediatric population.

Introduction

Primary pulmonary cancers in children are extremely rare. The International Agency for Research on Cancer announced that approximately 439 new primary pulmonary cancer cases were diagnosed in pediatric patients worldwide, accounting for fewer than 0.3% of all cancers diagnosed in patients between0 and 14 years of age and less than 0.02% of all lung cancers [1]. Mucoepidermoid carcinoma (MEC) often affects the salivary glands, representing the second most common salivary gland tumor [2]. MEC of the lung is an extremely rare neoplasm that accounts for <1% of all lung carcinomas [3]. The clinical symptoms and signs of pulmonary MEC include cough, hemoptysis, bronchitis, wheezing, fever, and chest pain, and commonly resemble the symptoms of pneumonia [4]. The prognosis of a localized low-grade MEC is better than that for high-grade tumors, and patients with low-grade

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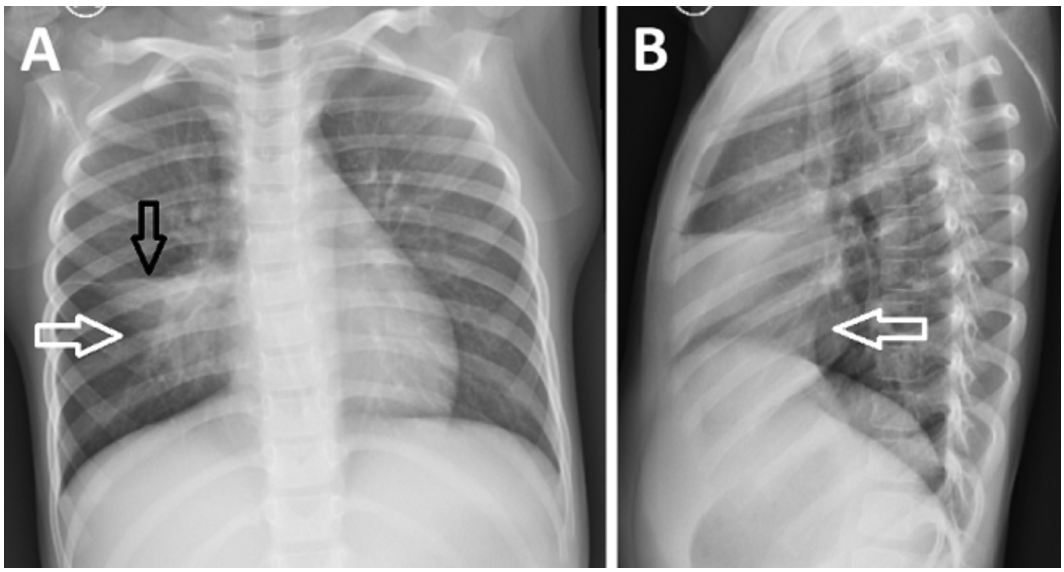

Fig. 1 – A chest X-ray at the time of admission showed consolidation in the right middle lobe (A and B, white arrows), which outlined the inferior margin of the horizontal fissure (A and B, black arrow).

MEC have a10-year overall survival rate of up to 72% [3]. In this article, we emphasized the clinical approach used to diagnose MEC and highlight the role played by chest computed tomography (CT) imaging for identifying the clues that led to the accurate diagnosis of this disease.

Case report

A 5-year-old girl presented to the hospital with pain in the right chest and a cough lasting for approximately1 month. The physical examination showed that the patient had a slightly elevated temperature (38°C) and a dull pain in the region of the right chest, near the heart.

A chest X-ray showed consolidation in the right middle lobe, and the consolidation outlined the inferior margin of the horizontal fissure (Fig. 1). The right hemi-diaphragm remained visible, and the right heart border had an indistinct appearance. The blood test showed slightly increased levels of neutrophils (12 G/L) and C-reactive protein (6 mg/L). The patient was treated with cefotaxime (1000 g every 12 hours) and linezolid (200 mg every 8 hours). However, after 1 month of treatment, the symptoms showed no improvement, and the chest X-ray findings remained unchanged (Fig. 2). A chest CT revealed a mass in the middle lobe of the right lung, which was homogeneous with well-defined borders (Fig. 2). The mass caused the occlusion of the right middle lobe bronchus, and the residual lung parenchyma of this lobe was necrotic (Fig. 2). A tumor in the right lung was suspected, and the patient underwent a tumor biopsy. The histological results revealed tumor cells that included epidermoid, mucous, and intermediate cells without keratinization. Based on these findings, an MEC was suspected.

A right middle lobectomy and mediastinal lymph node dissection were performed. The post-operative histological results revealed a MEC tumor (Fig. 3) without lymph node metastasis; however, microscopic tumor cut-through showed positive margins. The patient was treated with cisplatin, cyclophosphamide, and doxorubicin. After 2 cycles of chemotherapy, this patient has remained stable and the patient refused adjuvant chemotherapy. Chest CT scans showed no recurrence lesion (Fig. 4). The patient will be followed up through regular clinical examinations and chest CT scans.

Discussion

MEC is a rare malignant tumor of the lung, and the age of presentation ranges from 3 to 78 years [4]. No associations between smoking or other risk factors and MEC occurrence have been reported, and MEC does not appear to have any sex predilection [5,6]. MECs are typically endobronchial, often arising in the segmental or lobar bronchi [7]. Patients with MEC may present with the following symptoms: cough, expectoration, hemoptysis, fever, wheezing, dyspnea, and recurrent pneumonia. Additionally, some patients are asymptomatic [4,7]. The patient, in this case, presented with nonspecific symptoms; therefore, the diagnosis of tumor was easily missed.

Chest X-ray findings for MEC can include a pulmonary nodule or mass and signs of bronchial obstruction, such as pulmonary abscess, atelectasis, or post-obstructive pneumonia [8]. On CT scans, the mass may appear within the bronchus or in a peripheral pulmonary nodule, and the borders can be smooth, rounded, or lobulated [7,8]. Calcifications can be present, and low-grade tumors often enhance homogeneously; by contrast, heterogeneous enhancement suggests high-grade MEC [8]. Complications can include atelectasis, obstructive pneumonia, and pleural effusion [7]. Lung tumors

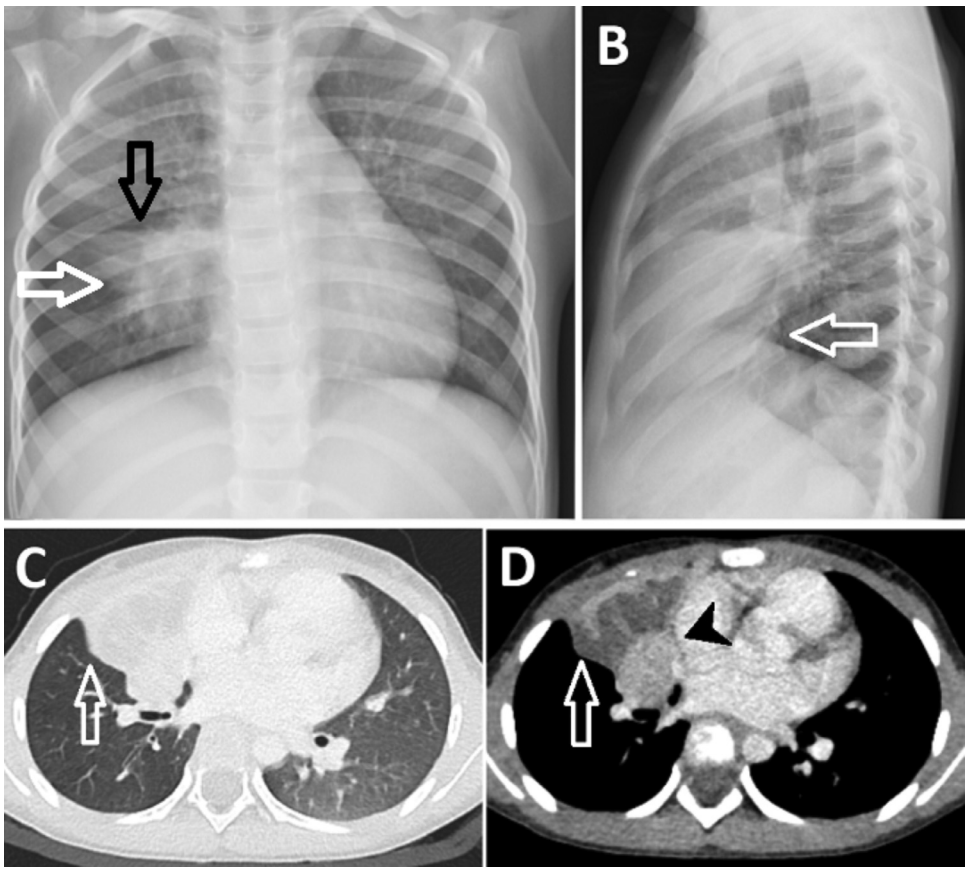

Fig. 2 – The chest X-ray after 1 month of antibiotic treatment showed that the lesion was constant (A and B, arrows). Chest computed tomography (CT) scans showed opacity in the middle lobe of the right lung (C, arrow), and the mass was well-circumscribed and homogeneous (D, arrowhead). The right middle lobe parenchyma was necrotic (D, arrow).

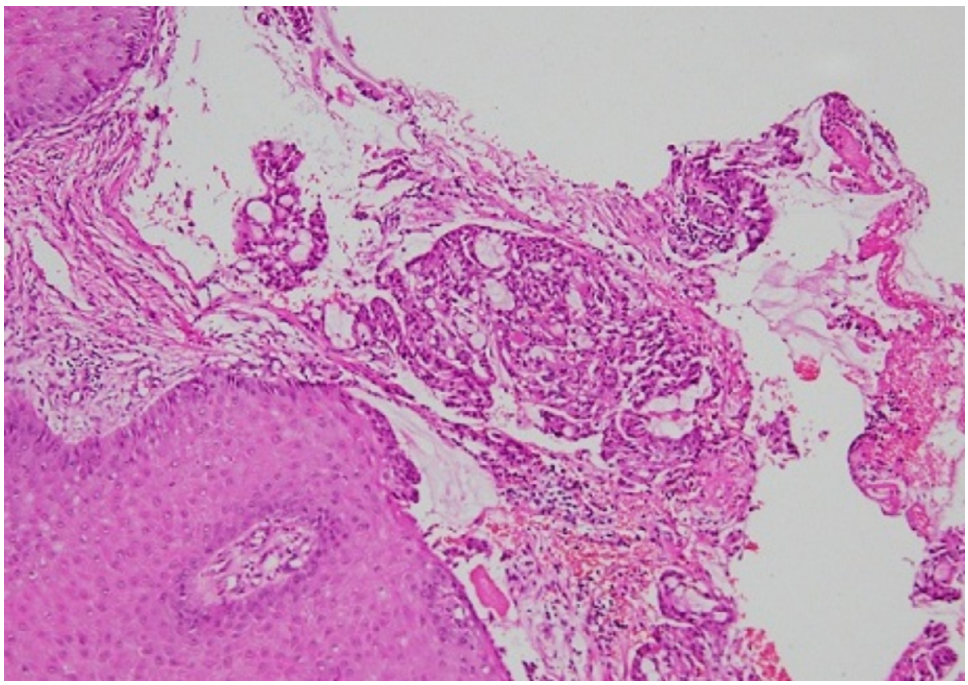

Fig. 3 – Hematoxylin and eosin stain of the tumor specimen (× 100) showed a mixture of epidermoid, mucous, and intermediate cells.

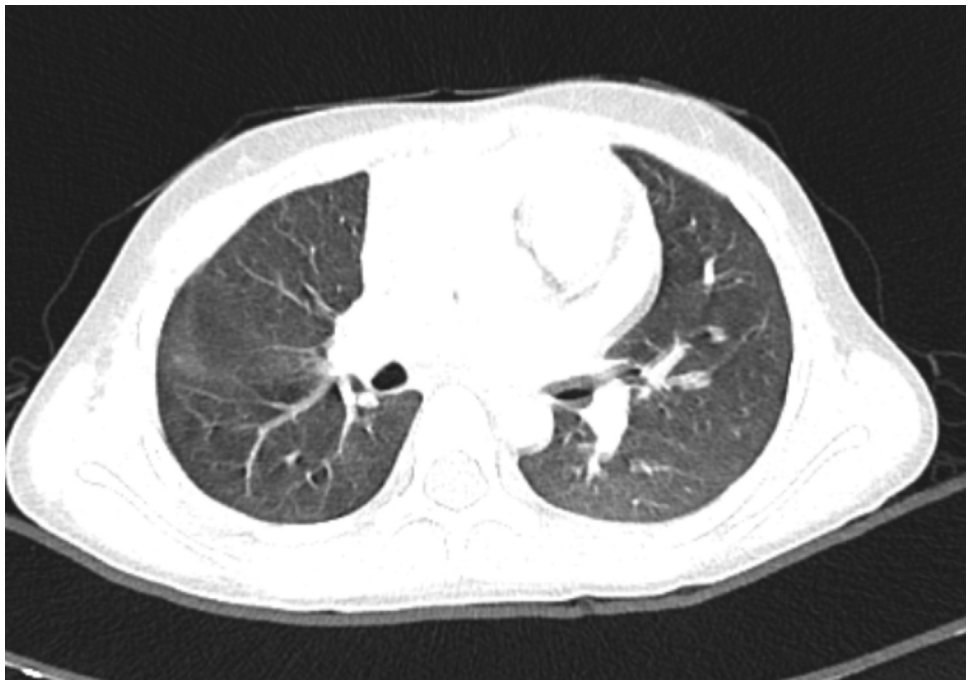

Fig. 4 – Chest CT scan after 1 month follow-up showed no recurrence lesion.

should always be included in the differential diagnosis for cases of recurrent pneumonia, especially when pneumonia repeatedly recurs in the same lung region [9]. In this patient, the chest X-ray showed right middle pneumonia; however, the clinical symptoms and CT results predicted a tumor, which was confirmed by the histological results.

Histologically, the tumor cells included variable proportions of epidermoid, mucous, and intermediate cells, with the absence of keratinization or *in situ* carcinoma of the surface epithelium [10]. The diagnosis of MEC was based on the histopathological examination due to the lack of specific clinical and radiological features [7].

Complete surgical resection is the optimal treatment for low-grade tumors, and adjuvant chemotherapy and radiation therapy are not typically indicated for cases associated with successful complete resection [11]. Patients who harbor specific endothelial growth factor receptor (EGFR) mutations can be treated with EGFR-targeted therapy [11]. In the cases of lymph node metastasis or positive surgical margins, and high-grade MEC, adjunct therapy such as radiotherapy and chemotherapy could be used [6,12,13]. However, there were no guidelines of adjuvant therapy for MEC. The 5-year survival rate associated with low-grade tumors (95%) is higher than that for high-grade (43%) [7].

For any pediatric patient who presents with fever and cough, pulmonary pneumonia should be excluded during the differential diagnosis at the time of admission. Chest X-ray findings at the first time of admission indicated a diagnosis of pneumonia or atelectasis. Commonly, antibiotic therapy is effective in these patients. The patient did not present during the COVID-19 pandemic, so it was not supposed due to COVID-19 pneumonia. Other causes might include other types of lung tumor. Although the MEC identified in this patient was low-grade, the complete tumor resection was not possible; therefore, the patient received adjuvant chemotherapy after surgery. This patient must be carefully followed up regularly.

Conclusion

In conclusion, MEC is a malignant tumor that rarely occurs in the lung. The clinical presentation and chest X-ray findings associated with MEC can sometimes resemble those for pneumonia; however, CT imaging features can suggest a tumor. When the lesion on CT suggests a tumor that is associated with persistent pneumonia that is unresponsive to antibiotic treatment, MEC should be included in the differential diagnoses, even in pediatric cases.

Ethical statement

Appropriate written informed consent was obtained for the publication of this case report and accompanying images.

Author contributions

Tran TT and Le TV contributed equally to this article as co-first authors. All authors have read the manuscript and agree to the contents.

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