Mucoepidermoid carcinoma: A case report

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April 09, 2024

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Keywords

pediatric pulmonology, cancer, lungs

1 INTRODUCTION

Primary lung neoplasms in children are rare. A significantly larger number of lung neoplasms are secondary neoplasms, mostly metastases from some other primary tumor process.[1] Primary lung tumors in children, although rare, are mostly malignant (75% of cases). Carcinoids account for 40% of these tumors, bronchogenic carcinomas for 17% of cases, and pleuropulmonary blastomas for about 15% of cases.[2] Mucoepidermoid carcinoma is the most common type of salivary gland carcinoma in the adult population. It can also be found in the bronchi and in the thyroid gland. It is not frequently found in the lungs, especially in children, where it constitutes approximately 0.1-0.2% of all primary lung tumors.[3]It originates from glandular tissue identical to that of the salivary glands, which is located in the submucosa of the trachea and bronchi.[4] We will present a rare case of mucoepidermoid carcinoma of the right bronchus in a six-year-old girl who was hospitalized due to right-sided pneumonia and pleural effusion.

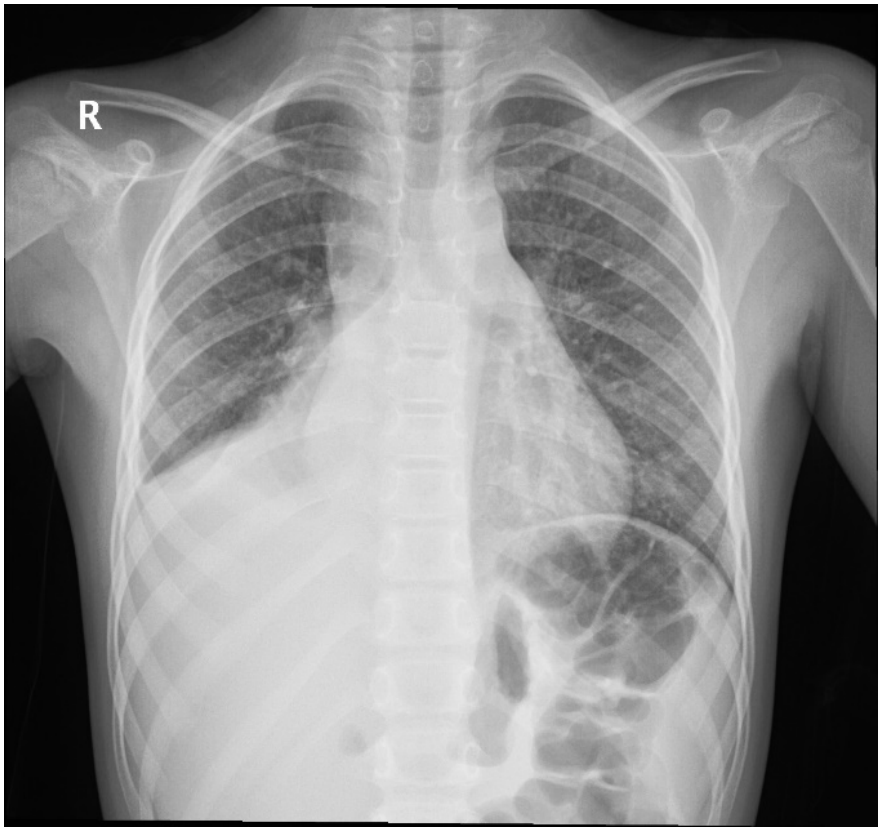
2 CASE PRESENTATION

A six-year-old patient was referred to our clinic due to an elevated body temperature, shivering, and vomiting. Laboratory diagnostics were performed at the local Health Center (CRP 338 mg/L, WBC 18.8 x 10^9/L (neutrophils 78%)), along with a chest X-ray, which revealed right-sided pneumonia with pleural effusion. The patient had previously experienced chickenpox seven months ago and had two episodes of pneumonia since, which were treated on an outpatient basis.

The clinical examination of the patient revealed the following: Subfebrile temperature (37.8°C), tachycardia (118/min); reduced breath sounds on auscultation over the right lung, with no breath sounds heard at the base. Additional radiological assessment (ultrasound of the lung base) confirmed the presence of pleural effusion.

Parenteral (ceftriaxone, clindamycin) and oral (azithromycin) antimicrobial therapy was prescribed, and a pediatric surgeon was consulted. There was no indication for pleural drainage.

The patient responded positively to the prescribed therapy, becoming afebrile on the fifth day of hospitalization. Follow-up X-rays showed partial regression of the previously described inflammatory changes, and a follow-up ultrasound confirmed the regression of pleural effusion. The girl was discharged for home treatment with continued oral antimicrobial therapy (cefpodoxime), with a scheduled follow-up appointment at the clinic.

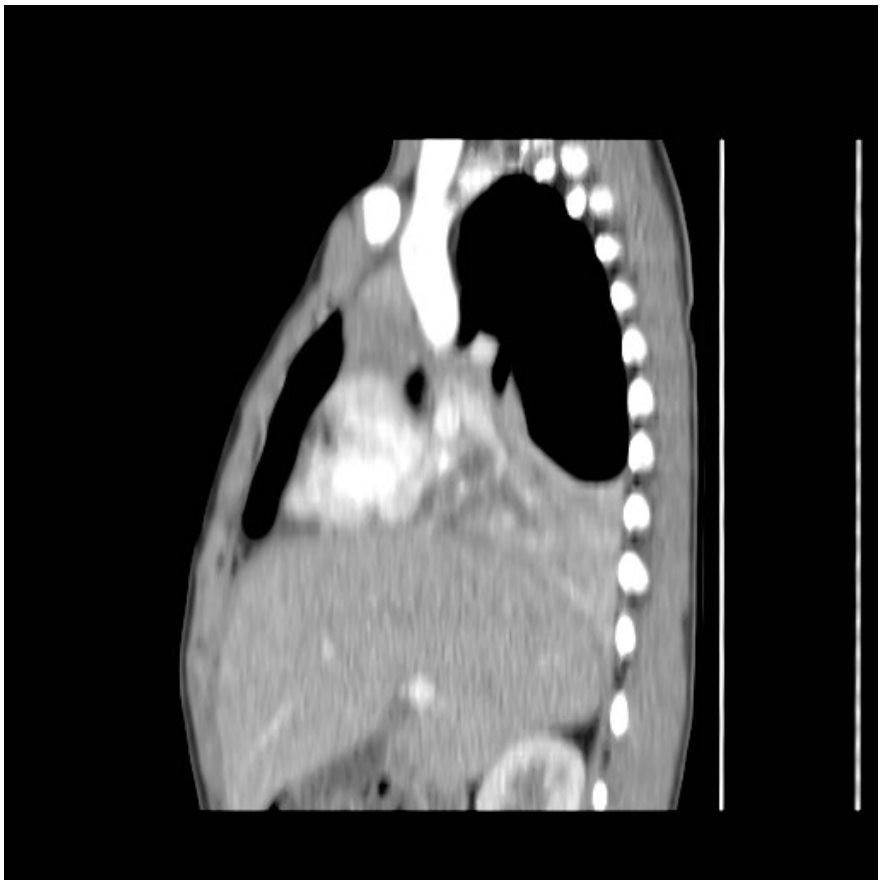


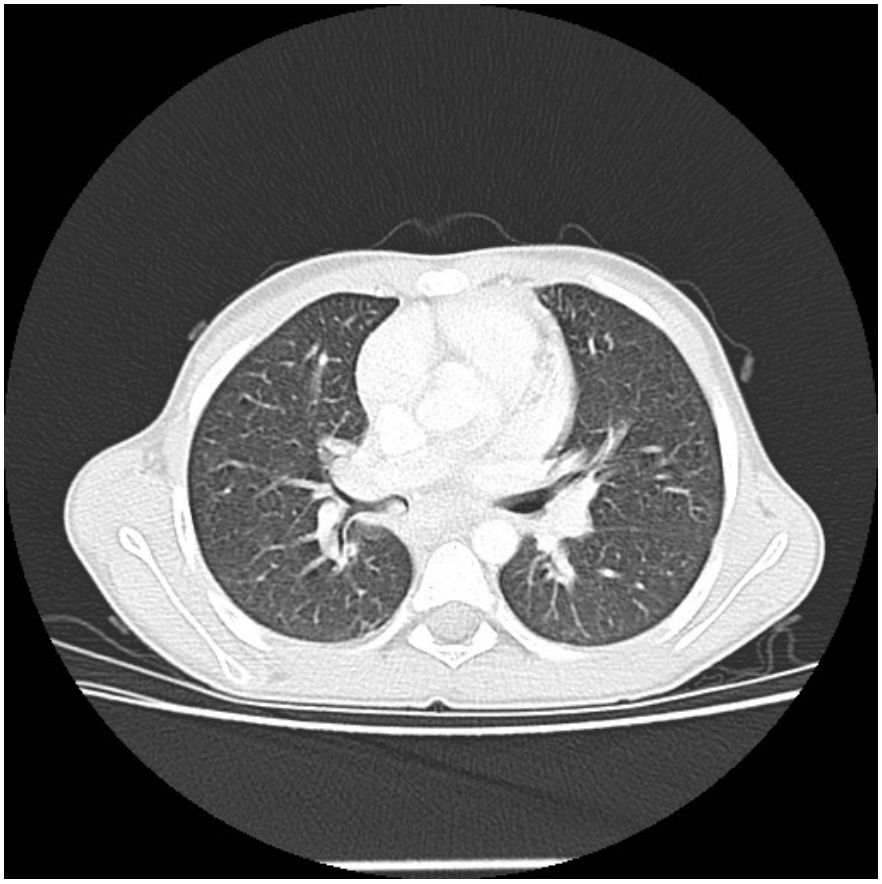
Two months later, the patient was readmitted due to right-sided pneumonia. On admission, she had a subfebrile temperature and appeared pale. There was reduced breath sounds over the right basal lung, and her abdomen was tender to palpation.

Laboratory diagnostics revealed a white blood cell count of 20.4 x 10^9/L (neutrophils 76%) and a CRP level of 293.7 mg/L. No pathogenic microorganisms were isolated in microbiological tests. A chest X-ray

showed persistent infiltrates on the right, with a suspicion of atelectasis of the middle lobe of the right lung. An ultrasound of the lung base revealed pleural effusion.

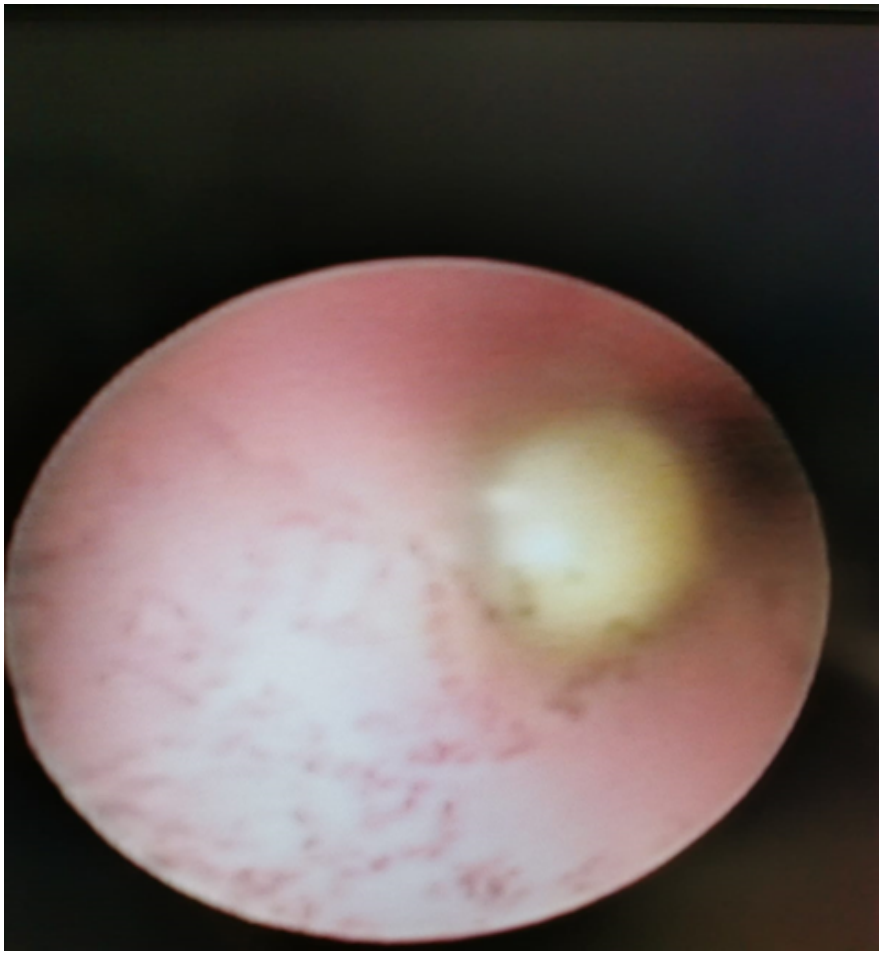
Empirical parenteral and oral antimicrobial therapy was prescribed. Due to the persistence of the infiltrates, a chest MSCT (multi-slice computrized tomography) was performed. The MSCT revealed heterogeneous content protruding into the right main bronchus, as well as the intermedius bronchus, and was spherical in shape (possible mucous content, food aspiration). There were also smaller conglomerates of lymph nodes in the mediastinum and right hilum.

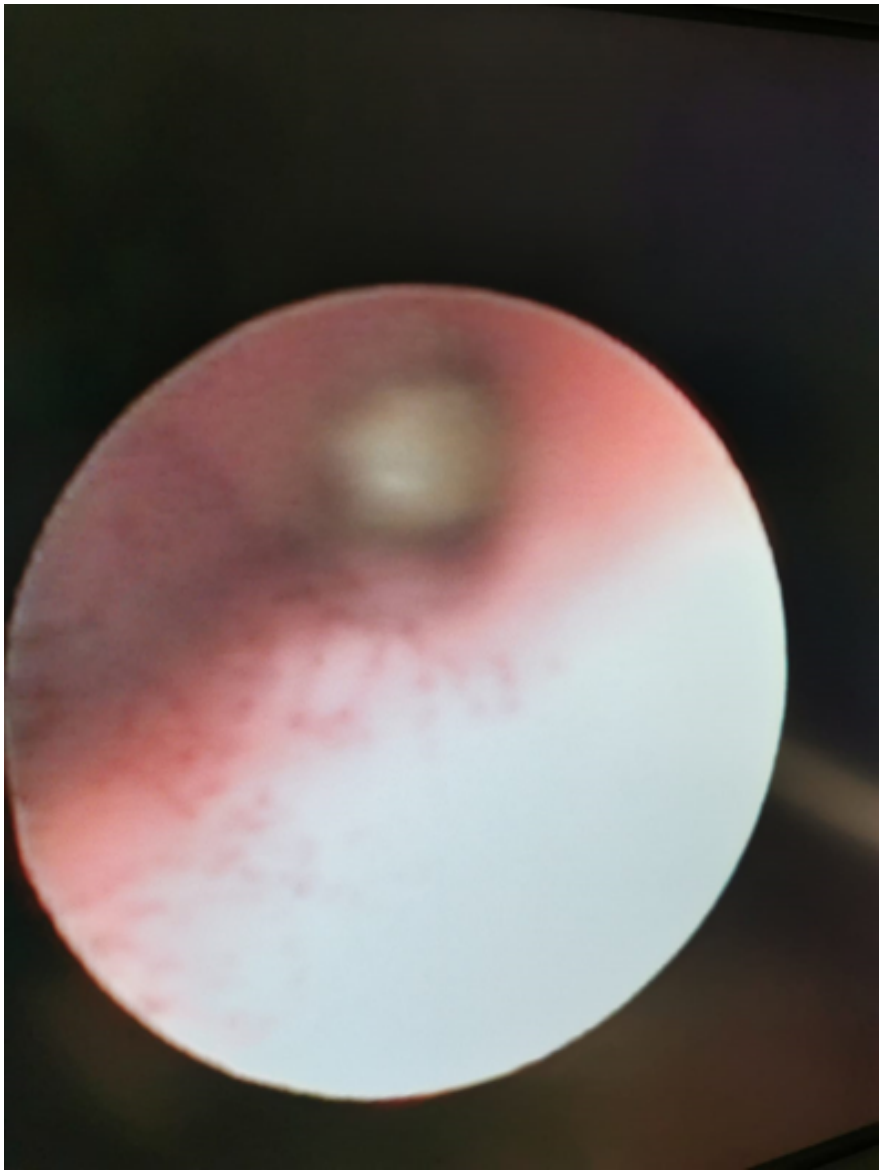




Considering the findings of the chest MSCT, a flexible bronchoscopy was performed. The entrance to the right intermediate bronchus was completely obstructed by a yellowish-white formation (possible mucous plug, foreign body). The formation protruded into the right main bronchus.

A recommendation was made for rigid bronchoscopy, which was performed under general anesthesia. A formation measuring 1.5 cm in length and up to 0.5 cm in thickness was extracted. A sample was sent for pathological analysis (PHD analysis).





Based on the PHD analysis, the biopsy material revealed fragments of cylindrical mucinous epithelium, metaplastic squamous epithelium, and an abundant amorphous pink substance (possible necrotic tissue) surrounded by evident granulation tissue with areas of suppurative inflammation.

Immunohistochemistry was applied using CK5/6, AE1/AE3, CD56, and Ki67.

It was decided to send the sample for further analysis to the Institute of Pathology and Cytology at KBC Zagreb-Rebro. A follow-up flexible bronchoscopy was performed, which indicated that the entrance to the right intermediate bronchus remained obstructed by a compact yellowish-white formation. The decision was made to postpone the rigid bronchoscopy until the PHD results were available.

The patient was afebrile for several days, and follow-up laboratory tests showed a decrease in inflammatory markers. However, there was another episode of fever, along with an increase in inflammatory markers. A combination antimicrobial therapy (clindamycin and meropenem) was prescribed, and the patient responded positively.

Since the initial biopsy material was insufficient for a correct analysis, it was decided to perform a rigid bronchoscopy and PHD material analysis at another medical center. The patient was referred to "Sestre milosrdnice" University Hospital Center in Zagreb.

Upon hospitalization at the new facility, the previously prescribed antimicrobial therapy was continued. A flexible bronchoscopy was performed, revealing a round, fibrous, and elastic formation in the right segmental bronchus, which filled the entire segmental bronchus (middle lobe on the right). Three samples were taken for analysis.

The PHD analysis results were consistent with mucoepidermoid carcinoma. The patient was transferred to the Clinic for Children's Diseases "Klaićeva" in Zagreb for further treatment.

Under general anesthesia, a right-sided thoracotomy was performed in the 5th intercostal space. The intermediate bronchus was resected, and an inferior bilobectomy was performed. Macroscopically, the tumor did not extend to the resection margin (sample number 1). Paratracheal lymph node dissection was carried out (sample number 2). A chest tube was placed under negative pressure.

After the procedure, the patient was transferred to the Intensive Care Unit. An ultrasound examination of the lungs confirmed complete consolidation of the upper lobe parenchyma and atelectasis on the right side. A flexible bronchoscopy was performed, and the right bronchus entrance was cleaned.

The patient's condition gradually improved, and the chest tube was removed on the sixth postoperative day. The subsequent course of the patient's stay was uneventful, and follow-up laboratory results were normal. The margins of the PHD preparations were clean, and there were no tumors in the lymph nodes; reactive changes were observed.

3 DISCUSSION

Primary lung tumors in children are rare, and they are most often malignant in nature.

Mucoepidermoid carcinoma (MEC) of the bronchus accounts for about 10% of primary lung tumors in the pediatric population.[5]

MEC primarily affects the large airways, leading to common symptoms such as cough, shortness of breath, fever, wheezing, chest pain, and hemoptysis.[6]

Radiological diagnostics may not be sufficient to establish a definitive diagnosis, and endoscopic examination, such as flexible bronchoscopy, is often required.

Chest X-rays are typically normal in cases where MEC is located in the tracheal region, but most bronchial lesions are characterized by lobar infiltrates, atelectasis, or bronchiectasis due to partial or complete endobronchial obstruction.[7]

Recurrent lung infections, especially in the same region, should raise suspicion of a possible underlying tumorous process.

MEC is often presented as an intraluminal mass leading to luminal occlusion.[8]

Metastasis to regional lymph nodes is rare in this type of tumor.

Due to the low metastatic potential of MEC, surgical treatment is usually the first option.[6]

The case discussed involved a right-sided thoracotomy in the 5th intercostal space, resection of the intermediate bronchus, and an inferior bilobectomy. Paratracheal lymph node dissection was performed.

Despite the rarity of this tumor, it's important to consider it in cases of persistent respiratory symptoms, especially when recurrent inflammatory processes occur in the same location.

The discussion emphasizes the importance of early recognition, accurate diagnosis, and appropriate surgical intervention in cases of mucoepidermoid carcinoma of the bronchus in pediatric patients.

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