CASE REPORT

REVISED Case Report: Giant lung hamartoma : An usual cause of lobectomy in a five-year child

[version 2; peer review: 1 approved, 2 approved with reservations]

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Abstract

Pulmonary hamartomas are the most common benign tumors of the lung in adults. They are usually asymptomatic because of their small size and their slow-growing character. We report the case of a 5-year-old child presenting with a giant lung mass causing recurrent right pneumonia. Surgical resection with middle lobectomy was performed. Final histology revealed pulmonary hamartoma with predominant adenofibromatous and lipomatous differentiation.

Keywords

lung tumor, case report, children, surgical intervention, hamartoma

Open Peer Review

Approval Status ✓ ? ?

	1	2	3
version 2 (revision) 24 Sep 2024	✓ view		? view
version 1 17 Jun 2024	✗ view	? view	

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Any reports and responses or comments on the article can be found at the end of the article.

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Author roles: Louhaichi S: Conceptualization, Data Curation, Investigation, Software, Writing – Original Draft Preparation, Writing – Review & Editing; **Hamdi B**: Conceptualization, Data Curation, Investigation, Writing – Original Draft Preparation, Writing – Review & Editing; **Bouacida I**: Data Curation, Investigation, Writing – Original Draft Preparation; **Haddar Y**: Data Curation, Investigation, Writing – Original Draft Preparation; **Trimech S**: Data Curation, Investigation; **Ammar J**: Data Curation, Supervision, Validation; **Ayadi A**: Data Curation, Formal Analysis, Investigation, Supervision, Writing – Review & Editing; **Hamzaoui A**: Investigation, Supervision, Validation, Writing – Review & Editing; **Ines B**: Formal Analysis, Supervision; **Marghli A**: Supervision

Competing interests: No competing interests were disclosed.

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REVISED **Amendments from Version 1**

More details related to surgery were added to give a clear and complete picture of the management of this benign tumor in children.

Any further responses from the reviewers can be found at the end of the article

Introduction

Lung hamartomas are benign pulmonary tumors characterized by an incidental finding in most cases.[1] Compocased of a mixture of variant mesenchymal elements, it is more frequently seen in male adults. Pediatric cases are extremely rare.[2,3] Herein we report the case of a pulmonary hamartoma revealed by persistent pneumonia in a five-year-old child.

Case report

A five-year-old child was referred to our department in September 2023 because an abnormal pulmonary density of the lower right hemithorax. His past medical history revealed recurrent admissions for right pneumonia during the last two years. The patient complained of chronic productive cough without chest pain or hemoptysis. Physical examination did not reveal abnormalities apart from a decrease in breath sounds in the right lung. Chest radiography revealed a heterogeneous right opacity above the diaphragm as showed in Figure 1.

Chest computed tomography revealed a giant cystic and solid mass measuring 122 × 80 × 102 mm compressing the right middle and lower lobes. This mass contained tissular, fatty, and calcified elements, along with multiple airy cysts, suggesting a giant pulmonary hamartoma (Figure 2).

Therapeutic options were discussed in a multidisciplinary reunion and surgery was decided. It was performed under general anaesthesia. The patient underwent a right lateral thoracotomy. During exploration, the mass occupied two-thirds of the right thorax and compressed the upper and lower lobes (Figure 3) as well as the mediastinum. It was carefully mobilized. It doesn't invade the mediastinum and the phrenic nerve was identified and preserved. The fatty mass depended on the middle lobe, which was a small strip of destroyed lung parenchyma. The surgical strategy was, to begin with an atypical resection removing the bloc of the mass followed by a complete right middle lobectomy. The dissection of the middle lobe arteries was challenging due to the destroyed tissue. The anatomical resection was successfully achieved and the patient was extubated immediately in the operating room. There were no anaesthetic complications during the procedure. The postoperative course was uneventful. The chest drain tube was removed three days postoperatively and the patient was discharged our days later.

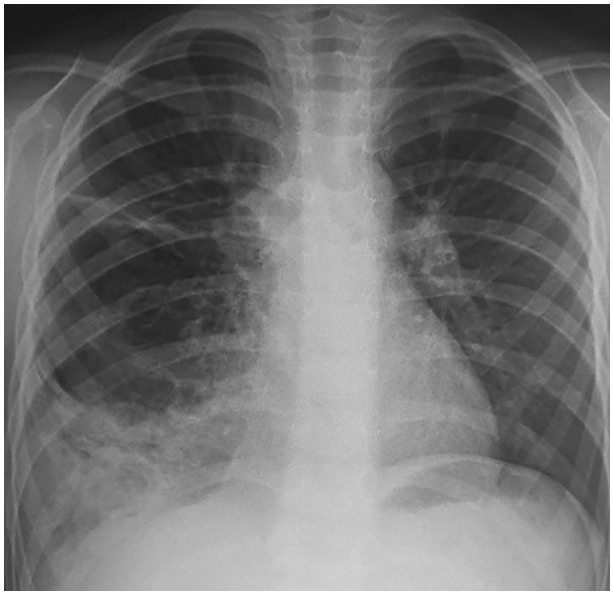

Figure 1. Chest radiograph view demonstrating a heterogeneous opacity in the middle and lower zones of the right lung.

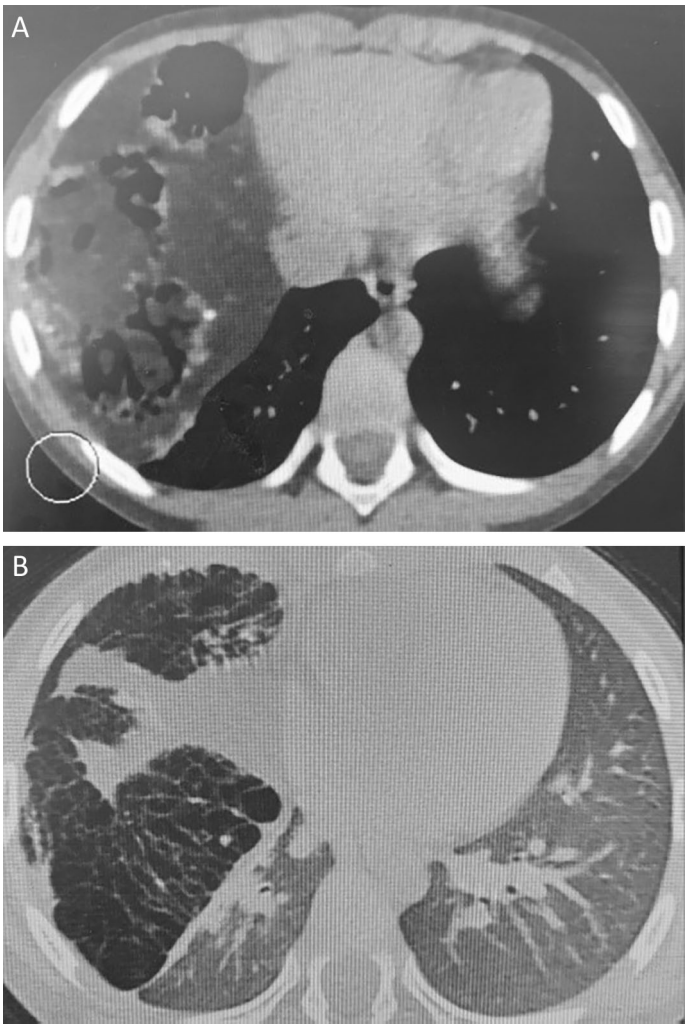

Figure 2. Pulmonary hamartoma: a contrast-enhanced lung CT scan is showing a large, lobulated soft-tissue density mass with foci of low attenuation and calcification with compression of the right lower lobe in mediastinal window (A) and lung window (B).

The final histology of the tumor showed a well-circumscribed mass measuring 14 × 10 × 5 cm, with predominantly adenofibromatous and lipomatous differentiation, calcifications, and ossified lesions (Figure 4). There was no evidence of cartilage or muscle tissue. The tumor was covered by a thin fibrous capsule. Additionally, diffuse alveolar hemorrhage lesions were observed in the middle lobe. Follow-up at three weeks post-surgery indicated no adverse outcomes.

Discussion

Pulmonary hamartomas are benign tumors that often occur in middle-aged adults with male predominance.[4] Within the pediatric population, pulmonary hamartomas are significantly rarer.[5] It is an incidental finding in most cases, with a diameter ranging from 1 to 8 cm.[6] This type of tumor has never been reported as a congenital lesion. Cytogenetic analysis showed abnormalities in chromosomal bands 6p21, 12q14–15, or other regions corresponding to mutations in high-mobility group (HMG) proteins. This group of proteins plays an important role in regulating chromatin architecture and gene expression.[7]

The pathological pattern of the tumor usually shows predominant chondroid differentiation with a mixture of adipose tissue, fibrous tissue, smooth muscle, and bone, along with entrapped respiratory epithelium**.** Immunohistochemical staining is not necessary for the diagnosis.[8] In other cases, the major component can define various subtypes of the tumor: lipomatous, adenoleiomyomatous, and fibrous hamartomas.[7,9] In the current case, the tumor consisted histologically of glandular lumens and fibrous tissue with some calcifications. No evidence of cartilage or muscle tissue damage was observed.

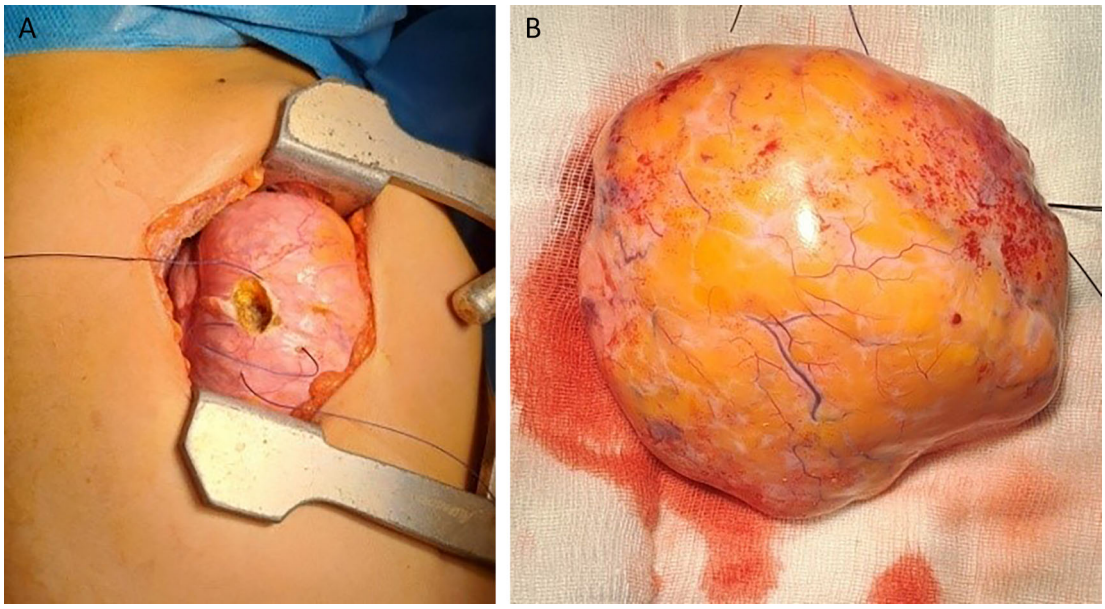

Figure 3. **Resection of the tumor (A). The hamartoma after resection (B).**

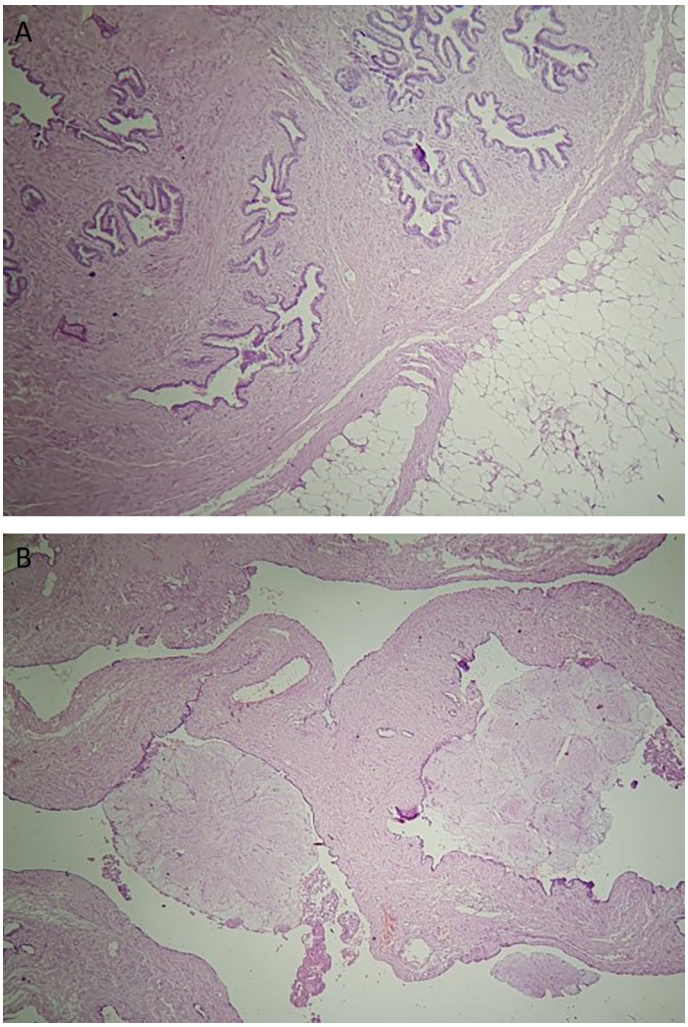

Figure 4. **HE*40 Pulmonary hamartoma with adenofibromatous proliferation associated adipose tissue (A) Adenofibromatous pattern with cystic changes and club-like papillae set against a collagenous stroma (B).**

Pulmonary hamartomas are typically asymptomatic. The patient had a medical history of recurrent pneumonia before being referred to our department. Respiratory infections may occur because of mechanical obstruction of the bronchus.

On tomodensitometry, lung hamartoma usually appears as a lobulated nodule with a heterogeneous density and no pleural traction. Characteristic imaging manifestations include the presence of fat (60% of the cases) and a popcorn appearance of calcifications observed in 5-50% of the cases.[10] Malignant transformation is exceedingly rare.[11]

Surgery is indicated for symptomatic masses or those in which malignancy cannot be excluded.[12,13] Enucleation and wedge resection are the most common surgical choices for preserving functional lung tissue.[14] However, in our case, tumor resection and middle lobectomy were mandatory because of the large size of the tumor and compression of the surrounding parenchyma.

Conclusions

Lung hamartomas typically occur in adults and are asymptomatic in most cases; parenchymal resection is rarely required when surgery is indicated. Our case is unusual because of its many peculiarities. A 5-year-old child presented with recurrent pneumonia. Moreover, owing to its large size, the tumor caused parenchymal damage, leading to middle lobectomy during surgery. Finally, the tumor was characterized by predominant adenofibromatous differentiation, with no cartilage. Knowledge of atypical presentations of this neoplasm is crucial to avoid misdiagnosis and to guide appropriate surgical treatment, especially in pediatric patients.

Consent

Written informed consent was obtained from the patient's parents for the publication of this case report and accompanying images.

Data availability

No data are associated with this article.

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Open Peer Review

Current Peer Review Status: ✔ ? ?

Version 2

Reviewer Report 04 March 2025

https://doi.org/10.5256/f1000research.171972.r361907

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Sevilay Özmen
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General Evaluation: This case report reports a rare presentation of a large pulmonary hamartoma in a pediatric patient. The article shows how the rare disease pulmonary hamartoma can present clinically in children. The authors have presented a successful case report with detailed information about the patient's history of recurrent pneumonia, imaging modalities, and surgical intervention. The article also describes such a rare case in the literature and sheds light on the diagnostic and therapeutic processes of similar cases.

The case highlights the atypical presentations of pulmonary hamartoma and how it may follow a different clinical course in pediatric patients. This may raise awareness in clinical practice.

It was stated that genetic analysis should be performed in some pulmonary hamartoma cases and some chromosomal abnormalities were observed. However, genetic analysis was not performed for this case and it would be useful to provide more information in this section.

“HMGA2 fusion in lung hamartomas suggests that these tumors share a similar pathogenesis with other mesenchymal tumors harboring HMGA2-related fusion genes (e.g., chondromas, extraskeletal osteochondromas, lipomas, and leiomyomas)” is stated in reference number 8. However, the source of information regarding molecular genetics is stated as number 7. This information needs to be corrected.

Is the background of the case’s history and progression described in sufficient detail?
Yes

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?
Yes

Is sufficient discussion included of the importance of the findings and their relevance to

future understanding of disease processes, diagnosis or treatment?
Yes

Is the case presented with sufficient detail to be useful for other practitioners?
Partly

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Pulmoner pathology, Hepatopankreato biliary pathology

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Reviewer Report 04 November 2024

https://doi.org/10.5256/f1000research.171972.r326621

Hüseyin Fatih Sezer
Department of Thoracic Surgery, Kocaeli University Faculty of Medicine, Kocaeli, Turkey

I reviewed the final version of the article.

Is the background of the case's history and progression described in sufficient detail?
Partly

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?
Partly

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?
Partly

Is the case presented with sufficient detail to be useful for other practitioners?
Partly

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: The article is a repetition of what is generally known and has no additional features, except for some surgical difficulty.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

Version 1

Reviewer Report 14 September 2024

https://doi.org/10.5256/f1000research.161126.r317823

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Kenneth J. Nobleza
[1] Baylor College of Medicine, Houston, Texas, USA
[2] Baylor College of Medicine, Houston, Texas, USA

Thank you for presenting this rare pulmonary hamartoma in children. Please find below a couple of comments you may want to consider for improving the manuscript:
- More details regarding the surgery (pre-, intra-, and post-op) would be needed.
- As the patient has got recurrent admission for right pneumonia for the past two years, serial results on chest radiography would be helpful.

Is the background of the case's history and progression described in sufficient detail?
Partly

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?
Yes

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?
Partly

Is the case presented with sufficient detail to be useful for other practitioners?
Partly

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Pediatrics

We confirm that we have read this submission and believe that we have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however we have significant reservations, as outlined above.

Reviewer Report 25 June 2024

https://doi.org/10.5256/f1000research.161126.r294003

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Dear Authors
First of all, I would like to thank you for the work you have done to prepare the article. My criticisms and suggestions about the article are as follows.
- The purpose of the article should be written more clearly in the introduction, and some more literature information should be added.
- The words 'case report, children, surgery' should be removed from keywords and more effective words should be added.
- In the discussion section, pathology is included too much, is the article a surgery article or a pathology article? In addition, information about surgery should be added to the discussion (e.g. post-operative complications, postoperative results, etc.) and why anatomical resection is performed or in which cases it can be performed should be explained more clearly and comprehensively.
-The conclusion section should be rewritten and unnecessary information should be removed. In addition, the discussion section should include the reasons why the case is considered unusual. Unfortunately, the article is a repetition of what is generally known and has no additional features, except for some surgical difficulty.

Is the background of the case's history and progression described in sufficient detail?
Partly

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?
Yes

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?
Partly

Is the case presented with sufficient detail to be useful for other practitioners?

Partly

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: The article is a repetition of what is generally known and has no additional features, except for some surgical difficulty.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to state that I do not consider it to be of an acceptable scientific standard, for reasons outlined above.