A child with multiple pulmonary hamartomas initially thought to be metastasis of Wilms' tumour

Bayram Altuntas[1]
Elif Demirci[2]
Zuhal Yildirim[3]
Yener Aydin[1]
Atilla Eroglu[1]

Author details can be found at the end of this article

Correspondence to:
Atilla Eroglu;
atilaeroglu@hotmail.com

Introduction

Hamartomas are the most common benign tumours of the lungs, comprising 8% of all lung tumours (Bini et al, 2002). These tumours usually appear as solitary pulmonary nodules on radiological imaging but they can also appear as multiple nodules. Establishing a differential diagnosis for patients with multiple hamartomas can be difficult. There are only 22 cases of multiple lung hamartomas in the literature, and only one case with multiple lung hamartomas that occurred after treatment for Wilms' tumour (Lindner and Willnow, 1987). This article presents a child who underwent left nephrectomy as part of treatment for Wilms' tumour and developed multiple hamartomas in both lungs afterwards.

Case report

A 5-year-old boy was admitted to the authors' clinic with multiple nodular lesions in both lungs which had been detected on a regular check up for a previous illness. Physical examination and laboratory test results were within normal limits. His past surgical history included a left nephrectomy 4 years ago as part of treatment for Wilms' tumour, and he had received chemotherapy postoperatively. A contrast-enhanced computed tomography scan of the chest revealed multiple nodular lesions in both lungs (Figure 1). These lesions did not show fludeoxyglucose uptake on positron emission tomography–computed tomography. The primary (Wilms') tumour was under control. With an initial diagnosis of pulmonary metastasis, ten nodules were surgically removed from the right lung via thoracotomy, and the two nodules in the left lung were removed using a thoracoscopic approach. Histopathological examination reported these to be adenofibrous hamartoma (Figure 2). The patient did not have any complications postoperatively.

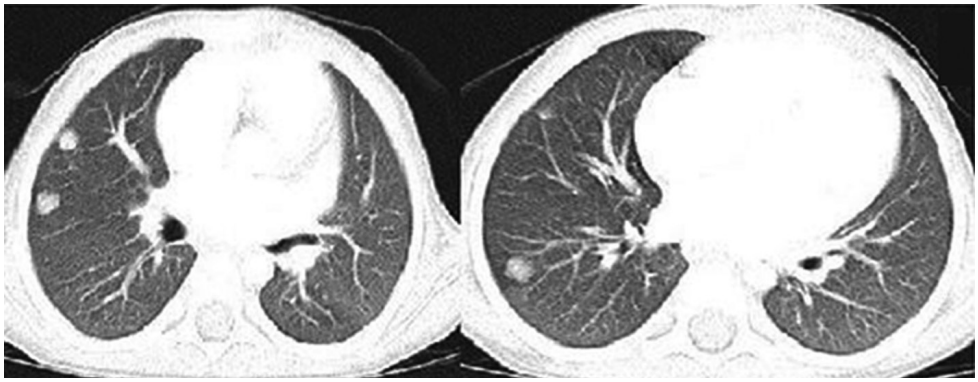

Figure 1. Computed tomography sections showing multiple lesions in both lungs.

Discussion

Pulmonary hamartomas are considered as true mesenchymal tumours and constitute 75% of all benign lung tumours (Teramoto and Suzumura, 2007). This patient did not have any pulmonary symptoms and his lesions were discovered at routine check-up. It is easy to diagnose these lesions preoperatively if they appear as solitary, calcified lesions with clear contours, localised peripherally on the radiological examination. However, establishing a diagnosis is more difficult when the lesions are multiple and localised endobronchially. In this case, preoperative radiological examinations revealed multiple, non-calcified lesions with partially clear contours, and the occurrence of these lesions after treatment for Wilms' tumour made the surgical team consider this to be metastasis.

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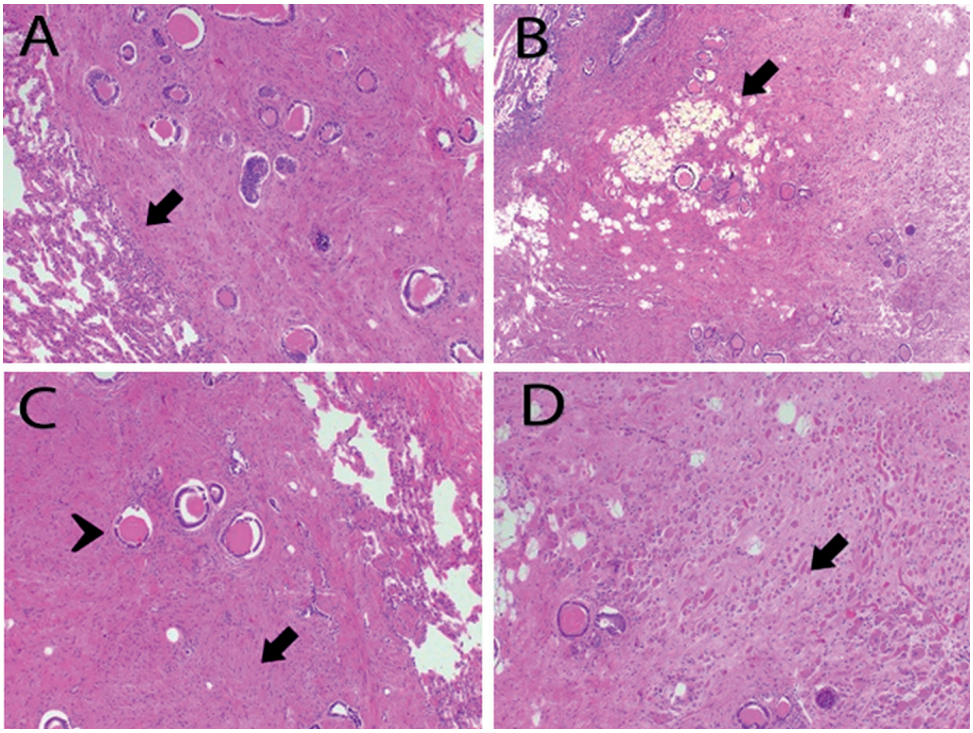

Figure 2. a. Alveolar structures (arrow) adjacent to mass. b. Mature adipose tissue (arrow). c. Mature glandular epithelial areas (arrowhead), intense fibroblastic ground (arrow). d. Mature muscle tissue.

Simple excision of these tumours is usually enough but lesions that localise endobronchially may require larger resection (Nistal et al, 2003). Multiple and deep parenchymal lesions are treated with thoracotomy, although a thoracoscopic approach might be applied for suitable cases. For this case, resection by thoracotomy was used for the right lung and thoracoscopic resection for the left lung.

Cowden disease and Carney's triad have been reported as forms of multiple hamartoma syndromes, but the condition of this patient was not consistent with either of these diagnoses.

The differential diagnosis for pulmonary hamartomas includes other coin lesions. Definitive histopathological diagnosis of these lesions is made after resection. It was possible that this patient's lesions were initially metastatic but later progressed to necrosis and afterwards fibrosis because he received chemotherapy following nephrectomy. However, histopathological examination did not reveal either necrosis or tumoural lesions; instead, it reported signs of adenofibrous hamartoma.

There are no precise physiopathological data available regarding the co-occurrence of Wilms' tumour and multiple pulmonary hamartomas. However, Lindner and Willnow (1987) reported that genetic factors might play a role in the development of Wilms' tumour and pulmonary hamartomas.

Pulmonary hamartomas are frequently accompanied by other developmental abnormalities and benign tumours, but the association of Wilms' tumour and pulmonary hamartomas has not been reported in the literature. With further prospective and experimental studies, the physiopathology of this relationship can be fully explained.

When multiple metastatic lesions of the lung are encountered, hamartomas must be kept in mind and the thoracoscopic approach applied for suitable cases.

Author details

[1]Department of Thoracic Surgery, Medical Faculty, Ataturk University, Erzurum, Turkey

[2]Department of Pathology, Medical Faculty, Ataturk University, Erzurum, Turkey

[3]Department of Pediatrics, Medical Faculty, Ataturk University, Erzurum, Turkey

Learning points

- After a patient has been treated for Wilms' tumour, the lung should be evaluated radiologically.
- When nodules resembling metastasis are detected, hamartomas should be kept in mind.

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