The hidden disease; bronchopulmonary sequestration as retrocardiac mass: a case report

Tugce Uzar[1], Mustafa Berk Sarac[2], Yağmur Saritas[1], Ozge Aksoy[3], Pankina Ekaterina[4], Adem Dirican[5], Sevket Ozkaya[6*]

European Journal of Medical Case Reports
Volume 3(1):08-11
© EJMCR. https://www.ejmcr.com/
Reprints and permissions:
https://www.discoverpublish.com/
https://doi.org/10.24911/ejmcr/173-1534019522

ABSTRACT

Background: Bronchopulmonary sequestration, simply called as pulmonary sequestration, is a relatively rare congenital anomaly with few reports of initial diagnosis occurring during adulthood. It consists of a nonfunctioning mass of lung tissue that lacks normal communication with the tracheobronchial tree and that receives its arterial blood supply from the systemic circulation.

Case Presentation: We report two cases with bronchopulmonary sequestration and a short review of the clinical/radiologic features of bronchopulmonary sequestration.

Conclusion: Look carefully at the retrocardiac area of chest radiography in adults for the definition of unexpected lesions. It can be bronchopulmonary sequestrations. It can be diagnosed easily with a feeding artery by an aberrant systemic artery which usually arises from the aorta.

Keywords: Bronchopulmonary sequestration, radiology, diagnosis.

Received: 24 August 2018 **Accepted:** 4 November 2018

Type of Article: CASE REPORT

Funding: None

Declaration of conflicting interests: None

Correspondence Author: Sevket Ozkaya
*Department of Pulmonary Medicine, Faculty of Medicine, Bahcesehir University, Istanbul, Turkey.
Email: ozkayasevket@yahoo.com
Full list of author information is available at the end of the article.

Background

Bronchopulmonary sequestration is a rare congenital malformation and it is characterized by a mass of non-functioning lung tissue separated from the normal bronchopulmonary tree and that receives its arterial blood supply from the systemic circulation. Although it is congenital, it is usually diagnosed in adults. It represents between 0.15% and 6.45% of all pulmonary malformations [1]. We report two cases with bronchopulmonary sequestration and a short review of the clinical/radiologic features of bronchopulmonary sequestration.

Case 1

A 19 years male patient was admitted to the hospital with complaints of a recurrent cough and fever. Chest radiograph showed reticular opacity on the retrocardiac area of the left hemithorax (Figure 1).

Thoracic computed tomography scan revealed a paravertebral mass lesion with air-space in the left lower lobe which was compatible with bronchopulmonary sequestration (Figure 2).

The patient eventually underwent a left thoracotomy with resection of the left mass lesion and it was consistent with the clinical/radiologic diagnosis of pulmonary sequestration.

Case 2

A 37 years male patient was admitted to hospital for check-up examination. There were no complaints and symptoms. Chest examination revealed the decreased breath sounds on the base of the left lower lung. Chest radiograph showed an abnormal opacity on the retrocardiac area of the left hemithorax (Figure 3).

Thoracic computed tomography scan revealed a paravertebral mass lesion in the left lower lobe (Figure 4).

Contrast-enhanced thoracic MRI and MR angiography were performed and there was an arterial vessel that arose directly from the lateral aspect of the aorta supplied the left lower lobe mass, consistent with a pulmonary sequestration (Figures 5 and 6). The patient did not accept any treatment.

Discussion

Pulmonary sequestration is a rare clinical condition and difficult to diagnose lung disease due to the absence of specific symptoms. The most common symptoms of pulmonary sequestration include recurrent episodes of fever, chest pain, and cough [2,3]. Radiologically, they usually escape from the eye due to the locations of the lesions, like as our cases. There are two subtypes: intralobar and extralobar, depending on whether it has independent pleura. An intralobar sequestration shares the visceral pleura of the adjacent lung, while the extralobar type is contained within its own visceral pleura, separate from the involved lung. Intralobar sequestration is more common (75% of cases) than the extralobar sequestration, roughly in a 3:1 ratio.

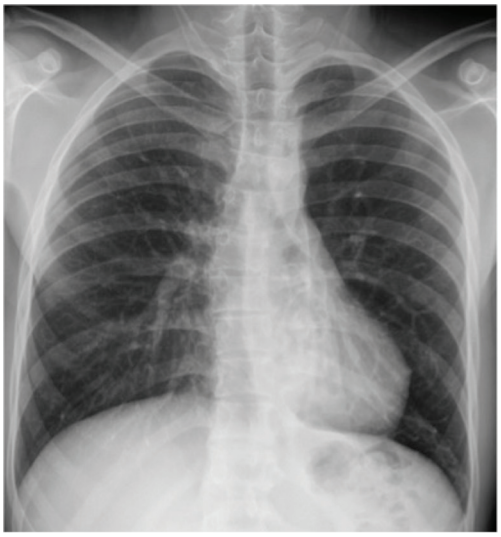

Figure 1. *Chest radiograph showing reticular opacity on the retrocardiac area of the left hemithorax.*

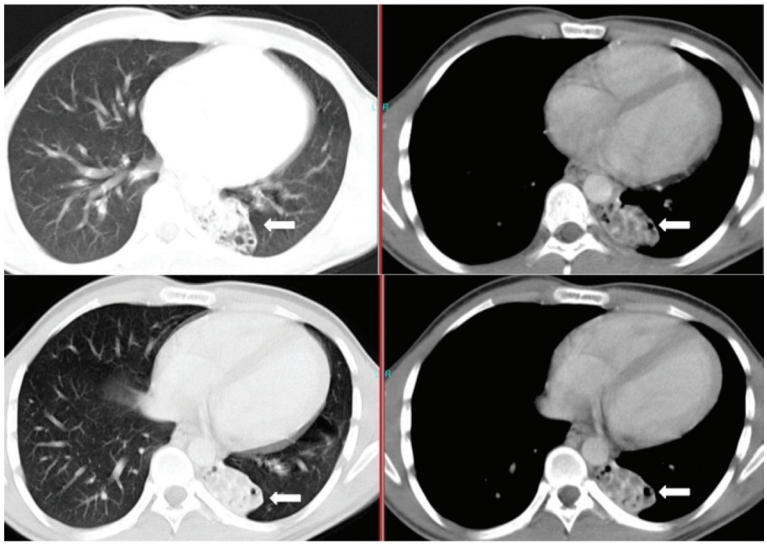

Figure 2. *Thoracic computed tomography scan showing a paravertebral mass lesion with air-space in the left lower lobe which was compatible with bronchopulmonary sequestration (Arrows).*

Extralobar disease is usually diagnosed in infancy or childhood, while intralobar disease often remains unrecognized until after the age of 20. Intralobar sequestration demonstrates no preferences for either lung, while extralobar sequestration is found in the left lung in 80% of cases [4–6]. The diagnosis is often suspected from the results of the clinical and radiological examination. The diagnostic investigation of choice remains aortography but it has to be demonstrated whether digital subtraction angiography or MRI angiography will prove more efficient and less invasive.

The most common diagnostic finding of bronchopulmonary sequestrations is a feeding artery by an aberrant systemic artery which usually arises from the aorta, as seen in our second case [7,8]. The mainstay of treatment has always been surgical excision. We recommend that the surgical treatment, usually complete excision in symptomatic patients.

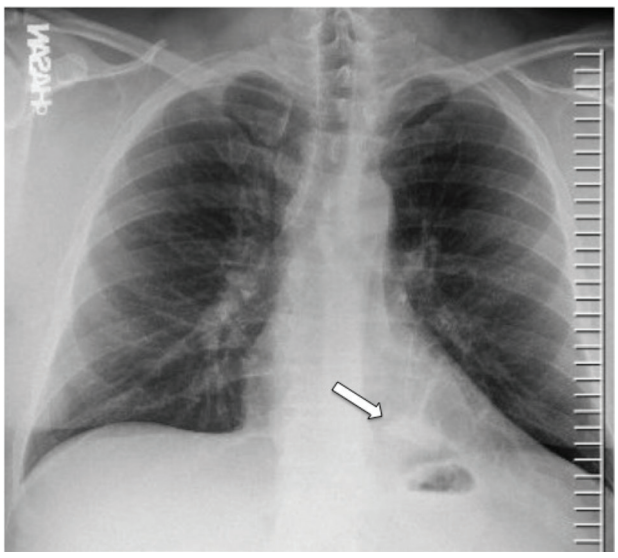

Figure 3. *Chest radiograph showing an abnormal opacity on the retrocardiac area of the left hemithorax (Arrow).*

Conclusion

In conclusion; look carefully at the retrocardiac area of the chest radiography in adults for the definition of unexpected lesions. It can be bronchopulmonary sequestrations. It can be diagnosed easily with a feeding artery by an aberrant systemic artery which usually arises from the aorta.

Acknowledgment

None.

List of Abbreviations

MRI	Magnetic Resonance Imaging
MR Angiography	Magnetic Resonance Angiography

Consent for publication

Written informed consents were obtained from the patients for publication of this manuscript and any accompanying images.

Ethical approval

Ethical approval is not required at our institution to publish an anonymous case report.

Author details

Tugce Uzar[1], Mustafa Berk Sarac[2], Yağmur Saritas[1], Ozge Aksoy[3], Pankina Ekaterina[4], Adem Dirican[5], Sevket Ozkaya[6]

1. Medical Student/Intern, Faculty of Medicine, Bahcesehir University, Istanbul, Turkey
2. Medical Student/Intern, Faculty of Medicine, Yeniyuzyil University, Istanbul, Turkey

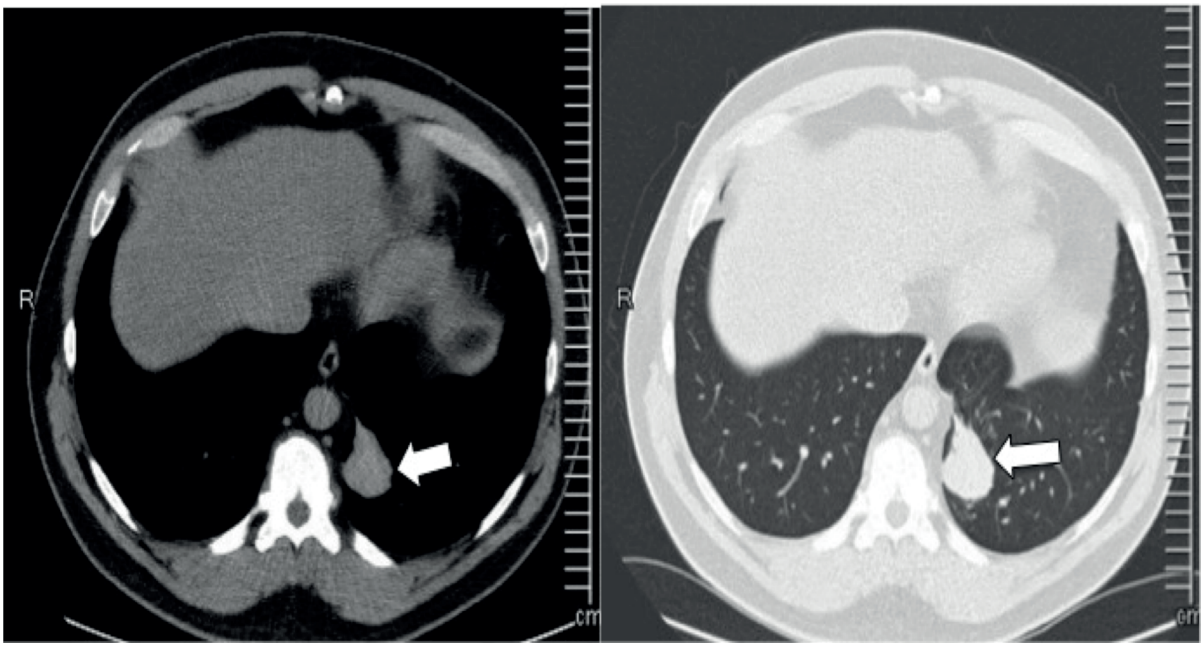

Figure 4. *Thoracic computed tomography scan showing a paravertebral mass lesion in the left lower lobe (Arrows).*

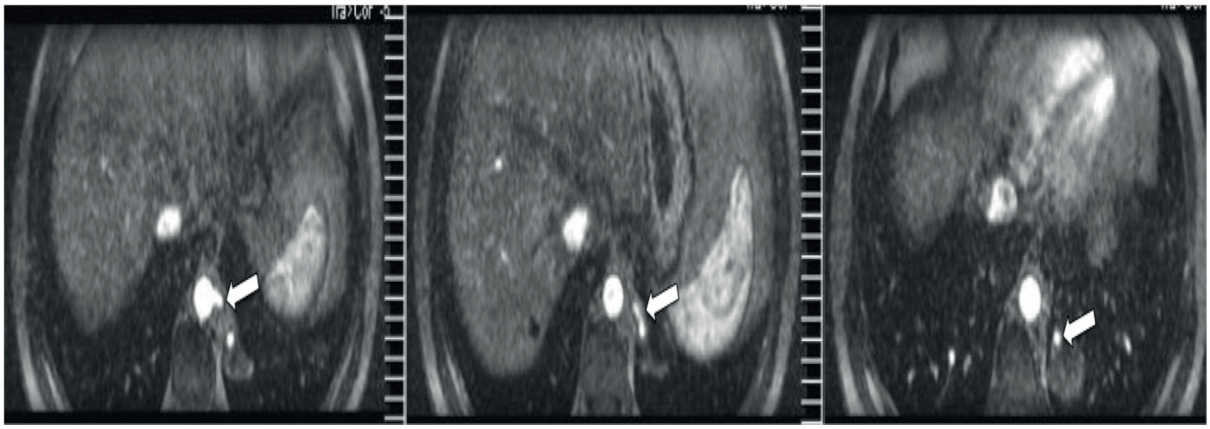

Figure 5. *Contrast-enhanced thoracic MRI showing an arterial vessel that arose directly from the lateral aspect of the aorta supplied the left lower lobe mass, consistent with a pulmonary sequestration (Arrows).*

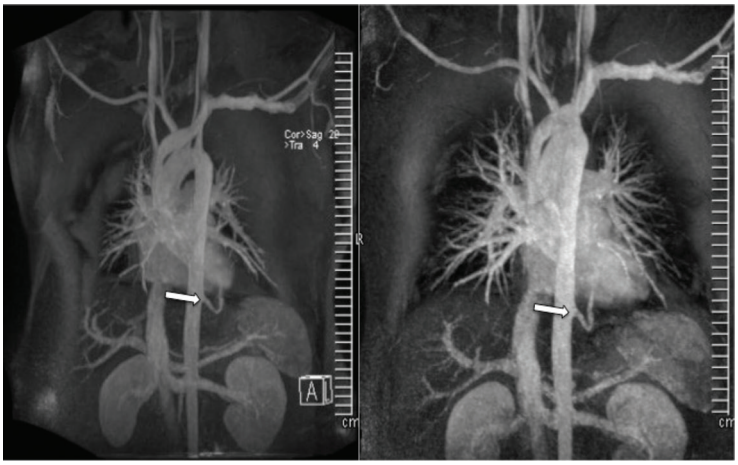

Figure 6. *Contrast-enhanced thoracic MR angiography was performed and there was an arterial vessel that arose directly from the lateral aspect of the aorta supplied the left lower lobe mass (Arrows).*

3. Medical Student/Intern, Faculty of Medicine, Kliment Ohridski University, Sofia, Bulgaria
4. Assistant, Department of Pulmonary Medicine, Faculty of Medicine, Bashkir State Medical University, Ufa, Russia
5. Pulmonologist, Department of Pulmonary Medicine, Samsun Medicalpark Hospital, Samsun, Turkey
6. Pulmonologist, Asociate Professor, Department of Pulmonary Medicine, Faculty of Medicine, Bahcesehir University, Istanbul, Turkey

References

1. Halkic N, Cue´noud PF, Corthe´sy ME. Pulmonary sequestration: a review of 26 cases. Euro J Cardio Thoracic Surg 1998; 14(2):127-33; https://doi.org/10.1016/S1010-7940(98)00154-7
2. Gustafson RA, Murray GF, Warden HE, Hill RC, Rozar GE. R.C. Intralobar sequestration a missed diagnosis. Ann Thorac Surg 1989; 47(6):841-7; https://doi.org/10.1016/0003-4975(89)90016-7
3. kamoto T, Masuya D, Nakashima T, Ishikawa S, Yamamoto Y, Huang CL, et al. Successful treatment for lung cancer associated with pulmonary sequestration. Ann Thorac Surg 2005; 80:2344-6; https://doi.org/10.1016/j.athoracsur.2004.06.065
4. Qian X, Sun Y, Liu D, Wu X, Wang Z, Tang Y. Pulmonary sequestration: a case report and literature review. Int J Clin Exp Med 2015; 8(11):21822–5.
5. Cooke CR. Bronchopulmonary sequestration. Respir Care 2006; 51:661-4.
6. Savic B, Birtel FJ, Tholen W, Funke HD, Knoche R. Lung sequestration: report of seven cases and review of 540 published cases. Thorax 1979; 34:96-101; https://doi.org/10.1136/thx.34.1.96
7. Jansen D, Schilte PM, De Graaff C, Van Dijk HA. Bronchopulmonary sequestration with an aneurysm of the aberrant artery. Ann Thorac Surg 1995; 60:193-4; https://doi.org/10.1016/S0003-4975(94)01065-X
8. Donovan CB, Edelman RR, Vrachliotis TG. Bronchopulmonary with MR angiographic evaluation. Angiology 1994; 45:239–44; https://doi.org/10.1177/000331979404500311

Summary of the case

Patient (gender, age)	1	Male, 19 year old; Male, 37 year old
Final diagnosis	2	Bronchopulmonary sequestration
Symptoms	3	Cough and fever
Medications	4	Surgery and follow-up
Clinical Procedure	5	Surgical resection
Specialty	6	Pulmonology and Radiology