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Pulmonary sequestration mimicking lung cancer – A case report

Plućni sekvestar koji imitira karcinom pluća

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Abstract

Introduction. Pulmonary sequestration is a rare congenital anomaly and most intralobar sequestrations were located in lower lobes. Case report. We reported an unusual 28-yearold female patient with intralobar pulmonary sequestration on the left lower lobe, successfully treated with lobectomy. Computed tomography (CT) of the chest with intravenous contrast revealed multiple clustered cystic lesions in the left lower lobe with aberrant artery from descedenting aorta. Additional aortography showed an aberrant artery (3 mm in diameter) arising from the abdominal aorta and flowing into the lesion. Conclusion. Standard therapy regimen for pulmonary sequestration includes surgery. CT scan of thorax with intravenous contrast and aortography represent the gold standard for its diagnosis. Tumor-like shadows seen on the chest radiography or CT scans should not be always suspected on malignant lesions.

Key words:

bronchopulmonary sequestration; diagnostic techniques and procedures; diagnosis, differential; thoracic surgical procedures.

Apstrakt

Uvod. Plućni sekvestar predstavlja retku kongenitalnu anomaliju, najčešće zastupljenu intralobarno u donjim režnjevima pluća. Prikaz bolesnika. Prikazali smo redak slučaj intralobarnog plućnog sekvestra kod bolesnice, stare 28 godina, uspešno operisane. Kompjuterska tomografija grudnog koša sa intravenskim kontrastom ukazala je na multiple cistične promene u levom donjem režnju, sa aberantnom arterijom iz descedentne aorte. Dodatno načinjena aortografija dokazala je aberentnu arteriju (3 mm u prečniku) iz abdominalne aorte koja se uliva u ledirano pluće. Zaključak. Standardni terapijski pristup za plućni sekvestar podrazumeva hirurško lečenje. Skener grudnog koša sa intravenskim kontrastom i arteriografija predstavljaju zlatne dijagnostičke standarde. Tumorske senke uočene na radiogramima grudnog koša ili na kompjuterizovanoj tomografiji ne moraju uvek da budu sumnjive na maligne promene.

Ključne reči:

sekvestracija, bronhopulmonalna; dijagnostičke tehnike i procedure; dijagnoza, diferencijalna; hirurgija, torakalna, procedure.

Introduction

Pulmonary sequestration is a rare congenital malformation of lung characterized by abnormal pulmonary parenchyma without tracheobronchial airway connection and with blood supply from a systemic artery ¹. Definitive treatment includes surgical excision, usually lobectomy, followed by division of the anomalous artery *via* standard thoracotomy.

Case report

A 28-year-old female was hospitalized due to chest pain and high temperature. Chest radiography showed a tumorlike shadow in the left lower lobe (Figure 1). Laboratory test findings showed elevated sedimentation rate – 94 mm/h, Creactive protein (CRP) – 198 mg/mL, blood cell count – 17.3 $\times 10^3$ /mm³ and D-dimer – 4.39 mg/L fibrinogen equivalent units (FEU). Electrocardiogram (ECG), heart ultrasound, lung functions tests and blood gas analysis were normal. A bronchoscopy finding was normal. The computed tomography (CT) scan of the chest with intravenous contrast revealed multiple clustered cystic lesions in the left lower lobe with the aberrant artery from the descedenting aorta (Figure 2). Aortography showed the aberrant artery (3 mm in diameter) arising from the abdominal aorta and flowing into the lesion (Figure 3). Left lower lobectomy was done. The intraoperative finding showed the abberant artery coming

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Fig. 2 – a) Thorax computed tomography (CT) scan: multicystic mass in the left lower lobe; b) Thorax CT scan: aberrant artery arises from the descedenting thoracic aorta.



Fig. 3 – Aortography showing the aberrant artery.

through the diaphragm into the mass (Figure 4). The artery was carefully dissected from the surrounding tissue. A pathological finding revealed intralobar pulmonary sequestration (Figure 5).

Discussion

Pulmonary sequestration is a rare congenital lesion of the lung parenchyma of unknown etiology without normal

connection with the tracheobronchial tree and with the blood supply directly from the descending aorta². It is a rare developmental abnormality which account for 0.15–6.4% of all congenital lung anomalies³. There are two morphological types: extralobar (25% of all cases), which have their own pleura, and intralobar (75% of all cases), which are surrounded by visceral pleura^{1,2}. Patients were presented in younger age with symptoms of recurrent bronchopulmonary infections and if they are recognized they should be operated on. If



Fig. 4 – Intraoperative photo showing the aberrant artery.



Fig. 5 – The left lower lobe with multicystic changes.

the symptoms of patients with pulmonary sequestration are not serious, intervention should be prolonged for several years. In the cases of repeated episodes of pneumonia, after physical investigation, diagnostic methods should include usual laboratory analysis, chest radiography and thorax CT scan with intravenous contrast. In our patient thorax CT scan with intravenous contrast revealed the aberrant artery from the descedenting aorta. Examination was completed with digital subtraction arteriography. Pulmonary sequestration is often misdiagnosed as congenital pulmonary cysts of bronchiectasis complicated with infection, benign lung tumor, diaphragmatic hernia and lung cancer⁴. Surgical resection is a definitive treatment in patients with pulmonary sequestration. Standard posterolateral thoracotomy has been used for a years. Because of its benign etiology partial lung resection for pulmonary sequestration should be more appropriate than lobectomy. Video assisted thoracoscopic surgery (VATS) is a better alternative to standard thoracotomy for pulmonary sequestration because of minimal surgical trauma, postoperative pain and duration of hospitalization ^{5, 6}. In the presented patient VATS was not done because of deficient technical equipment. Potential barriers to VATS include intrapleural adhesion due to the inflammatory process and dissection and dividing of the feeding artery, but rarely reported in case series ^{6, 7}. There are a few reports on successful management of asymptomatic pulmonary sequestration with angiographic embolization ⁸.

Conclusion

In a young patient tumor-like shadow on chest radiography should be suspected on a benign lesion. Detailed radiological investigations, including thorax CT scan, followed with arteriography, are mandatory.

REFERENCES

- 1. Okamoto J, Kubokura H, Usuda J. Safe transection of aberrant arteries associated with pulmonary sequestrations. BMC Surg 2015; 15(1): 27.
- Kestenbolz PB, Schneiter D, Hillinger S, Lardinois D, Weder W. Thoracoscopic treatment of pulmonary sequestration. Eur J Ccardiothorac Surg 2006; 29(5): 815–8.
- Sanic B, Birtel FJ, Tholen W, Funke HD, Knoche R. Lung sequestration: report of seven cases and review of 540 published cases. Thorax 1979; 34(1): 96–101.
- Ahmed M, Jacobi V, Vogl TJ. Multislice CT and CT angiography for non-invasive evaluation of bronchopulmonary sequestration. Eur Radiol 2004; 14(11): 2141–3.
- Avgerinos D, Reyes A, Plantilla E, Krikhely M. Video-assisted thoracoscopic surgery for intralobar pulmonary sequestration. Cases J 2008; 1(1): 269.

- 6. *Wan IY, Lee TW, Siboe AD, Ng CS, Yim AP.* Video-assisted thoracic surgery lobectomy for pulmonary sequestration. Ann Thorac Surg 2002; 73(2): 639–40.
- Hamaji M, Burt BM, Ali SO, Mirkovic J. An incidental and uncommon pulmonary sequestration with an uncommon feeding artery. Int J Surg Case Rep 2013; 4(10): 861–2.
- Seok JP, Kim YJ, Cho HM, Ryu HY. A rare case of bilateral pulmonary sequestration managed with embolization and surgical resection in a patient. Korean J Thorac Cardiovasc Surg 2013; 46(6): 475–7.

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