DOI: https://doi.org/10.2298/SARH180410070T
UDC: 616.24-002-06; 616.24-006-073

CASE REPORT / ПРИКАЗ БОЛЕСНИКА

Exogenous lipoid pneumonia mimicking multifocal subpleural tumors

Dragana Tegeltija[1,2], Aleksandra Lovrenski[1,2], Tijana Vasiljević[1,3], Golub Samardžija[1,4], Ivan Kuhajda[1,2]

[1]University of Novi Sad, Faculty of Medicine, Novi Sad, Serbia;
[2]Institute for Lung Diseases of Vojvodina, Sremska Kamenica, Serbia;
[3]Oncology Institute of Vojvodina, Sremska Kamenica, Serbia;
[4]Institute for Cardiovascular Diseases of Vojvodina, Sremska Kamenica, Serbia

SUMMARY

Introduction Exogenous lipoid pneumonia (ELP) is caused by inhalation or aspiration of different oily substances of animal, vegetable, or mineral origin. It can be in acute or chronic form. Herein, we report a case of ELP in its chronic form, confirmed in surgical lung biopsy.

Case outline A 47-year-old male locomotive engineer, former smoker, without clinical symptoms, with a history of pneumonia two years previously, was referred to our institution. The operating diagnosis of multifocal subpleural tumors was made based on the chest computed tomography. A surgical lung biopsy confirmed a diagnosis of ELP.

Conclusion Diagnosis of ELP is frequently made after surgical biopsy performed for suspected neoplasm, because of neglecting profesional exposure to mineral oils.

Keywords: exogenous lipoid pneumonia; machine oil; surgical biopsy

INTRODUCTION

Lipoid pneumonia is an uncommon lung disease caused by the presence of lipids in the alveoli. It is classified as exogenous or endogenous. The endogenous type occurs secondary due to pulmonary alveolar proteinosis, chronic pulmonary bacterial or fungal infections, lipid storage diseases, and bronchial obstruction by tumors or broncholithiasis. Exogenous lipoid pneumonia (ELP) type is associated with the inhalation or aspiration of different oils. The clinical symptoms and radiological findings of ELP are nonspecific, depending on the patient's age, the amount of oily substances, and the length of the inhalation or aspiration period [1–5]. Different pulmonary diseases can resemble ELP. The diagnosis of ELP is based on a history of exposure to oil and the presence of lipid-laden macrophages in sputum or bronchoalveolar lavage or histopathology specimens [1, 5, 6].

There are no standard protocols for the treatment of ELP, but recommendations include discontinuing exposure to the oily agent, oxygen therapy, lung lavage, systemic corticosteroids, and surgical resection of lung tissue unresponsive to medical treatment [1, 4, 5, 6]. Various complications of ELP that can be found in the literature [7].

In the text below, we report a case of chronic form ELP confirmed by surgical lung biopsy.

CASE REPORT

A 47-year-old male who worked as a locomotive engineer was referred to our institution for the evaluation of lung disease which was initially diagnosed as a multifocal subpleural tumor (lipoma or fibroma). The patient, who was a former smoker, had no clinical symptoms and his only medical condition was pneumonia diagnosed two years previously. Chest computed tomography (CT) was performed. Nodular masses were present, measuring 30 mm in the upper and 34 mm in the left middle lobe, with fat density (Figure 1). Bronchoscopy samples were nondiagnostic. Two CT scans and two bronchoscopies were performed afterwards, but the nature of the disease was not clarified and the patient was admitted to our institution.

A physical and cardiovascular examination and routine blood tests showed no abnormal findings. A high-resolution CT scan was performed and showed persistent radiological findings. At a consultative meeting, a decision was made to perform video-assisted thoracoscopic surgery (VATS). VATS was performed and lung biopsy from the middle lobe showed a nodule with cavitation (Figure 2). There were multinucleated giant cells and lipid-laden macrophages in the cavity wall. Chronic interstitial lymphoplasmacytic inflammation formed well-circumscribed aggregates around airways in multiple areas of bioptic sample (Figure 3). Additional immunohistochemical analysis (panCK, vimentin, CD68, EMA, CD1a, CD10, and S100) excluded malignant diseases and set the diagnosis of ELP (Figure 4).

Received • Примљено: April 10, 2018
Revised • Ревизија: April 10, 2019
Accepted • Прихваћено: May 20, 2019
Online first: June 19, 2019

Correspondence to:
Dragana TEGELTIJA
Put doktora Goldmana 4
21204 Sremska Kamenica, Serbia
dragana.tegeltija@mf.uns.ac.rs;
tegeltijadragana@gmail.com

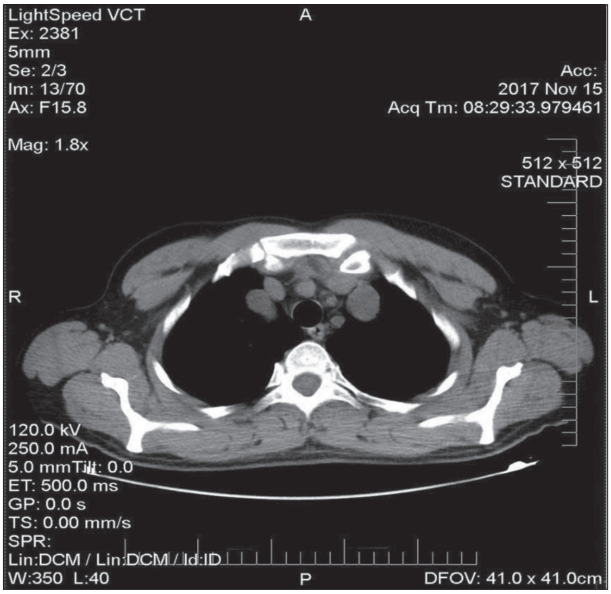

Figure 1. Chest computed tomography showing bilateral nodules

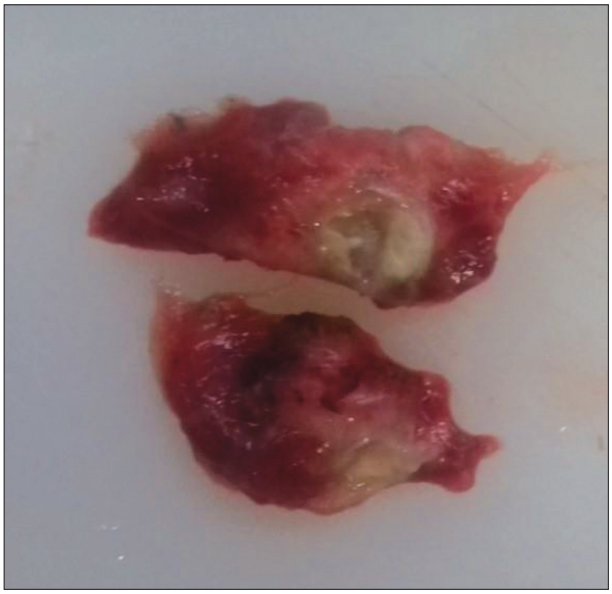

Figure 2. A node with a cavity in the surgical biopsy sample

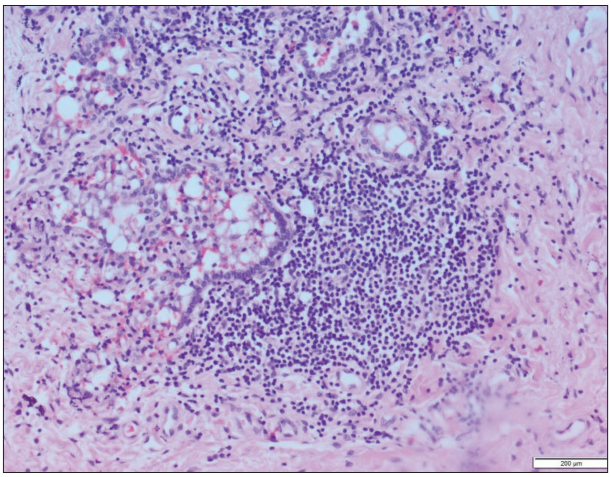

Figure 3. Chronic interstitial inflammation consisting of dense bronchocentric lymphoplasmacytic infiltrates (H&E, 10×)

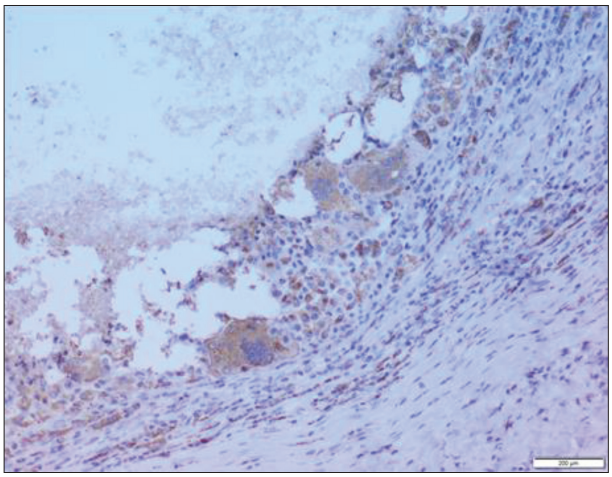

Figure 4. CD68 positivity in multinucleated giant cells (immunohistochemistry, 10×)

Six months after surgery, there was no radiological regression of other described lesions and pulmonary function tests were not modified.

This case report was approved by the institutional ethics committee, and written consent was obtained from the patient for the publication of this case report and any accompanying images.

DISCUSSION

ELP is the most common type of lipoid pneumonia and has been reported as a result of aspiration or inhalation of oil substances (animal, vegetal, or mineral origin) [4, 5, 6, 8]. In our case, no exogenous source was found initially. Once the histologic diagnosis was made, we asked the patient precisely if he had been exposed to mineral oils or had some risk factor for aspiration. The patient sad he had been working as a locomotive engineer for the last 20 years and had been in contact with mineral oil every working day. For this reason, we believed that in our case, ELP was the result of professional exposure to mineral oil.

ELP can be classified as acute or chronic. The acute form of ELP is caused by accidental aspiration of a large quantity of mineral oil in a short period of time. CT scan opacities are typically ground-glass or consolidative and can be seen in most patients within 24 hours [9, 10, 11]. The chronic form typically occurs in older patients with predisposing anatomic or functional abnormality in swallowing, but it has also been reported in children with cleft palate and mental retardation. The diagnosis of chronic form of ELP is set on average 38 days after the onset of nonspecific clinical symptoms such as cough, fever, weight loss, vomiting, and recurrent respiratory infections [12, 13, 14]. In contrast to other cases, our patient had no symptoms.

ELP may occur in all ages, most commonly in patients with gastroesophageal reflux, palpitations, swallowing dysfunction, and after administration of drugs [1, 2, 12, 13]. This opinion was confirmed by Sias et al. [1], who analyzed ELP formed as a consequence of the use of laxatives as a

result of intestinal obstruction due to Ascaris lumbricoides. This study involved 15 girls and 13 boys aged 1–108 months. In contrast to the above data, our patient was older, as in most published papers with individual ELP case reports [2, 3, 6].

Radiological changes are non-specific, mostly localized to the right lung [1, 3]. Changes seen on CT are also nonspecific and may be unilateral, bilateral and multifocal, consolidation with air bronchogram, crazy-paving, interlobular septal thickening, cavitation, and calcification [1, 14, 15, 16]. Jin et al. [4] analyzed 18 cases and found this change in 13 patients: geographical lobular distribution of ground-glass, miliary changes on both sides, interstitium thickening, cavitation, and mediastinal pneumatosis. The fact that it is difficult to set the ELP diagnosis to 18F-fluorodeoxyglucose positron-emission tomography is confirmed by the case of a patient suffering from Kaposi sarcoma. Two spicular changes (25 mm and 9 mm, respectively) in the upper right lobe showed fat density (-30–150 HU) on the CT scan, but the value of SUV 5 without local and distal expansion induced suspicion for a malignant tumor; therefore, lobectomy was multidisciplinary suggested and performed. Pathohistological examination confirmed ELP [8]. A differential diagnosis of ELP includes the following: nonspecific interstitial pneumonia, collagen vascular diseases, chronic eosinophilic pneumonia, idiopathic pulmonary fibrosis, hypersensitive pneumonitis, sarcoidosis, lung tumors (benign and malignant), bacterial pneumonia (acute and chronic), and pulmonary alveolar proteinosis [5, 17]. In our case, nodular lesions registered on CT were initially diagnosed as a bilateral subpleural lipoma or fibroma as their mean density was -30 HU.

ELP may be indicated by the following: data on aspiration or inhalation of oily substances, radiological findings and the presence of lipid-laden macrophages in the sputum, bronchoalveolar lavage, or histological sample [1, 3, 9]. Upon macroscopic examination, lung parenchyma is usually consolidated and yellowish stained, while cavitations are rare [18]. Pathohistological examination can show bronchocentric lymphoplasmacytic cell infiltration with multinucleated giant cells with cholesterol crystals in the cytoplasm, intraalveolar clumps of alveolar macrophages, giant cell granulomas, chronic inflammatory reaction, and interstitial fibrosis [5, 6]. Long-term exposure to an oily material can lead to the development of lung fibrosis with the destruction of normal parenchyma and the development of pulmonary heart, while bacterial superinfections and pulmonary aspergillosis are rare [16, 19]. In our case, ELP was confirmed on permanent paraffin sections and additional immunohistochemical analysis of a surgical sample obtained by VATS. We found a 15-mm-diameter nodule with the cavity filled with friable yellowish-white content and multinucleated giant cells and lipid-laden macrophages in the cavity wall.

The prevention of exposure to oily substances, supportive oxygenotherapy, multiple bronhoalveolar lavage, steroid therapy, and surgical resection represent several modalities of ELP treatment [1, 6, 7]. It was suggested to our patient to avoid machine oil, with a recommendation that the remaining nodular changes should be surgically removed after complete recovery, which he accepted.

Conflict of Interest: None declared.

REFERENCES

1. Sias SM, Ferreira AS, Daltro PA, Caetano RL, Moreira JS, Quirico-Santos T. Evolution of exogenous lipoid pneumonia in children: clinical aspects, radiological aspects and the role of bronchoalveolar lavage. J Bras Pneumol. 2009;35(9):839–45.
2. Filho APL. Exogenous lipoid pneumonia: importance of clinical history to the diagnosis. J Bras Pneumol. 2006;32(6):596–8.
3. Kuroyama M, Kagawa H, Kitada S, Maekura RM, Hirano H. Exogenous lipoid pneumonia caused by repeated sesame oil pulling: a report of two cases. BMC Pulm Med. 2015;15:135.
4. Jin YM, Ge LS, Ji W. Not typical pneumonia: acute exogenous lipoid pneumonia in 19 children. Int J Clin Exp Med. 2016;9(5):8557–62.
5. Rahaghi F, Varasteh A, Memarpour R, Tashtoush B. Teppanyaki/Hibachi pneumonitis: an exotic cause of exogenous lipoid pneumonia. Case Rep Pulmonol. 2016;2016:1035601.
6. Pielaszkiewicz-Wydra M, Bożena Homola-Piekarska B, Szcześniak E, Ciołek-Zdun M, Fall A. Exogenous lipoid pneumonia – a case report of a fire-eater. Pol J Radiol. 2012;77(4):60–4.
7. Ridaura-Sanz C, López-Corella E, Salazar-Flores M. Exogenous lipoid pneumonia superinfected with acid-fast bacilli in infants: a report of nine cases. Fetal Pediatr Pathol. 2006;25(2):107–17.
8. Chardin D, Nivaggioni G, Viau P, Butori C, Padovani B, Grangeaon C, et al. False positive 18FDG PET-CT results due to exogenous lipoid pneumonia secondary to oily drug inhalation: a case report. Medicine (Baltimore). 2017;96(22):e6889.
9. Lococo F, Cesario A, Porziella V, Mulè A, Petrone G, Margaritora S, et al. Idiopathic lipoid pneumonia successfully treated with prednisolone. Heart Lung J Crit Care. 2012;41(2):184–7.
10. Nakashima S, Ishimatsu Y, Hara S, Kitaichi M, Kohno S. Exogenous lipoid pneumonia successfully treated with bronchoscopic segmental lavage therapy. Respir Care. 2015;60(1):e1–5.
11. Indumathi CK, Vikram KS, Paul P, Lewin S. Severe lipoid pneumonia following aspiration of machine oil: successful treatment with steroids case report. Indian J Chest Dis Allied Sci. 2012;54(3):197–9.
12. García Latorre R, Rodríguez Díaz R, Barrios Barreto D, Ayala Carbonero A, García Gómez-Muriel MI, Gorospe Sarasúa L. Exogenous lipoid pneumonia in laryngectomy patients: radiological findings. Arch Bronconeumol. 2015;51(7):e36–9.
13. Baron SE, Haramati LB, Rivera VT. Radiological and clinical findings in acute and chronic exogenous lipoid pneumonia. J Thorac Imaging. 2003;18(4):217–24.
14. Simmons A, Rouf E, Whittle J. Not your thypical pneumonia: a case of exogenous lipoid pneumonia. J Gen Intern Med. 2007;22(11):1613–6.
15. Tavare AN, Creer D, Hayward M, Hare SS. Exogenous lipoid pneumonia mimicking multifocal bronchogenic carcinoma. J Thorac Oncol. 2015;10(12):1809–10.
16. Betancourt SL, Martinez-Jimenez S, Rossi SE, Truong MT, Carrillo J, Erasmus JJ. Lipoid pneumonia: spectrum of clinical and radiologic manifestations. AJR Am J Roentgenol. 2010;194(1):103–9.
17. Gondouin A, Manzoni P, Ranfaing E, Brun J, Cadranel J, Sadoun D, et al. Exogenous lipid pneumonia: a retrospective multicentre study of 44 cases in France. Eur Respir J. 1996;9(7):1463–9.
18. Gaerte SC, Meyer CA, Winer-Muram HT, Tarver RD, Conces DJ Jr. Fat-containin lesions of the chest. Radiographics. 2002;22 Spec No:S61–78.
19. Brown-Elliott BA, Griffith DE, Wallace RJ Jr. Newly described or emerging human species of nontuberculous mycobacteria. Infect Dis Clin North Am. 2002;16(1):187– 220.

Егзогена липоидна пнеумонија која опонаша мултифокалне субплеуралне туморе

Драгана Тегелтија[1,2], Александра Ловренски[1,2], Тијана Васиљевић[1,3], Голуб Самарџија[1,4], Иван Кухајда[1,2]

[1]Универзитет у Новом Саду, Медицински факултет, Нови Сад, Србија;
[2]Институт за плућне болести Војводине, Сремска Каменица, Србија;
[3]Институт за онкологију Војводине, Сремска Каменица, Србија;
[4]Институт за кардиоваскуларне болести Војводине, Сремска Каменица, Србија

САЖЕТАК

Увод Егзогена липоидна пнеумонија (ЕЛП) узрокована је инхалацијом или аспирацијом различитих уљаних супстанци животињског, биљног или минералног порекла. Може да буде акутна или хронична форма.

Приказујемо случај хроничне форме ЕЛП који је потврђен у хируршком биопсијском узорку.

Приказ болесника Мушкарац стар 47 година, машиновођа, бивши пушач, без клиничких симптома, две године после прележане пнеумоније јавио се у нашу установу. Радна дијагноза мултифокалних субплеуралних тумора је била заснована на компјутеризованој томографији грудног коша. Дијагноза ЕЛП је потврђена у хируршком биопсијском узорку.

Закључак Дијагноза ЕЛП се често поставља на хируршкој биопсији учињеној због сумње на неоплазму, услед занемаривања података о професионалној изложености минералним уљима.

Кључне речи: егзогена липоидна пнеумонија; машинско уље; хируршка биопсија