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Case Report / Приказ болесника

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Exogenous lipoid pneumonia mimicking multifocal subpleural tumors

Егзогена липоидна пнеумонија која опонаша мултифокалне субплеуралне туморе

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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 acute or chronic form.

Herein, we report a case of chronic form of ELP 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 ago referred in 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 in surgical biopsy performed for suspicion of neoplasm, because of neglecting profesional exposure to mineral oils.

Keywords: exogenous lipoid pneumonia; machine oil; surgical biopsy

САЖЕТАК

Увод Егзогена липоидна пнеумонија (ЕЛП) је узрокована 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 случаја Мушкарац стар 47 година, машиновођа, бивши пушач, без клиничких симптома две године након 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

INTRODUCTION

Lipoid pneumonia is an uncommon lung disease caused by the presence of the lipids in the alveoli. It is classified into endogenous and exogenous type. Endogenous type occurs secondary due to pulmonary alveolar proteinosis, chronic pulmonary bacterial or fungal infections, lipid storage diseases and the bronchial obstruction by tumors or broncholithiasis. Exogenous lipoid pneumonia (ELP) type is associated with inhalation or aspiration of different oils. The clinical symptoms and radiological findings ELP are nonspecific, depending on the patient's age, the amount of oily substances and the length of the inhalation or aspiration period [1, 2, 3, 4, 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 on sputum or bronchoalveolar lavage or histopathology specimens [1, 5, 6].

There are no standard protocols for 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 literature [7].

In the text below, we report a case chronic form of 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 evaluation of lung disease which was initially diagnosed as a multifocal subpleural tumor (lipoma or fibroma). The patient who was former smoker had no clinical symptoms and his only medical condition was pneumonia diagnosed two years ago. Chest computed tomography (CT) was performed. The 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. The high-resolution computed tomography (HR 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 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).

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

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 was exposed to some mineral oil or has some risk factor for aspiration. The patient sad he worked as a locomotive engineer for the last 20 years and has been in contact with mineral oil every working day. For this reason, we believe that in our cause ELP was the result of a professional exposure to mineral oil.

ELP can be classified into acute or chronic form. Acute form of ELP is caused by accidental aspiration of a large quantity of a 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 also has been reported in children with cleft palate and mental retardation. The chronic form ELP diagnosis 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. who analyzed ELP formed as a consequence of the use of laxatives due to intestinal obstruction due to Ascaris lumbricoides. This study involved 15 girls and 13 boys aged 1 to 108 months [1]. 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 non-specific and may be: unilateral, bilateral and multifocal, consolidation with air bronchogram, crazy-paving, interlobular septal thickening, cavitations and calcifications [1, 14, 15, 16]. Yi-Mei Jin 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 [4]. 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 (25.0 and 9.0 mm) 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 of a malignant

tumor and therefore lobectomy was multidisciplinary suggested and performed. Pathohistological examination confirmed ELP [8]. A differential diagnosis of ELP includes: 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 because their density was mean -30 Hounsfield units.

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 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 15.0 mm diameter nodule with cavity filled with friable yellowish-white content and multinucleated giant cells and lipid-laden macrophages in cavity wall.

The prevention of exposure to oily substances, supportive oxygenotherapy, multiple bronhoalveolar lavage, steroid therapy and surgical resection represent several modalities of treatment ELP [1, 6, 7]. Our patient was suggested 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.

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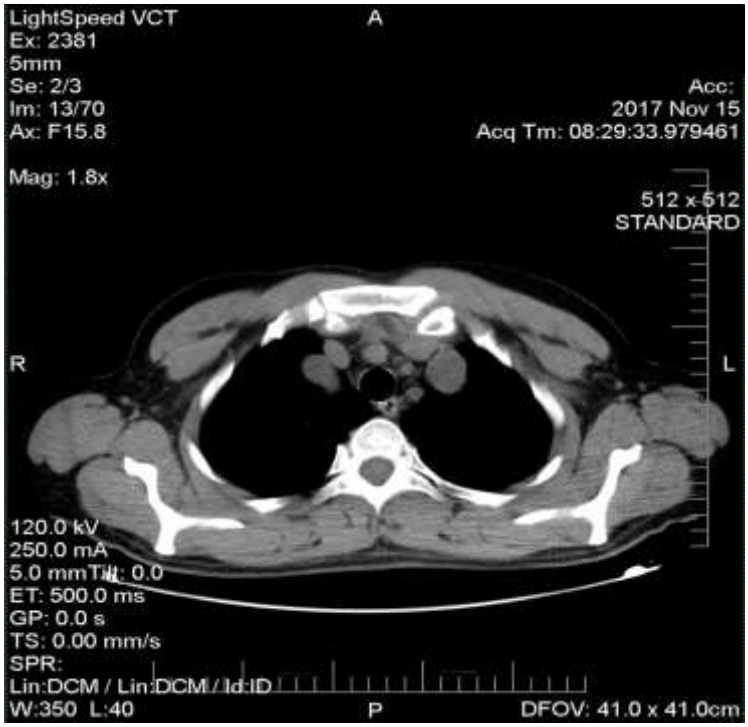

Figure 1. Chest computed tomography showing bilateral nodules

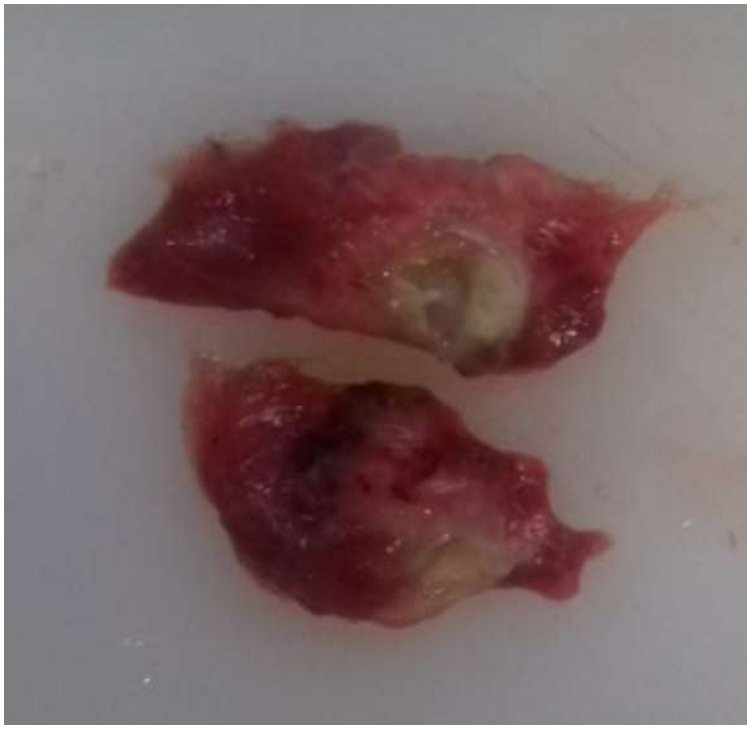

Figure 2. A node with cavity in surgical biopsy sample

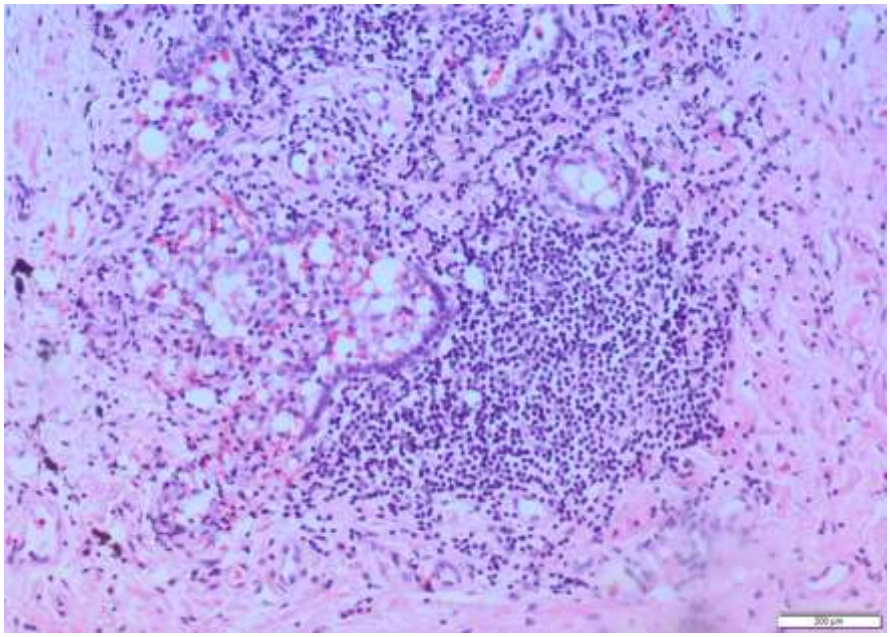

Figure 3. Chronic interstitial inflammation consisting of dense bronchocentric lymphoplasmocytic infiltrates (H&E, 10×)

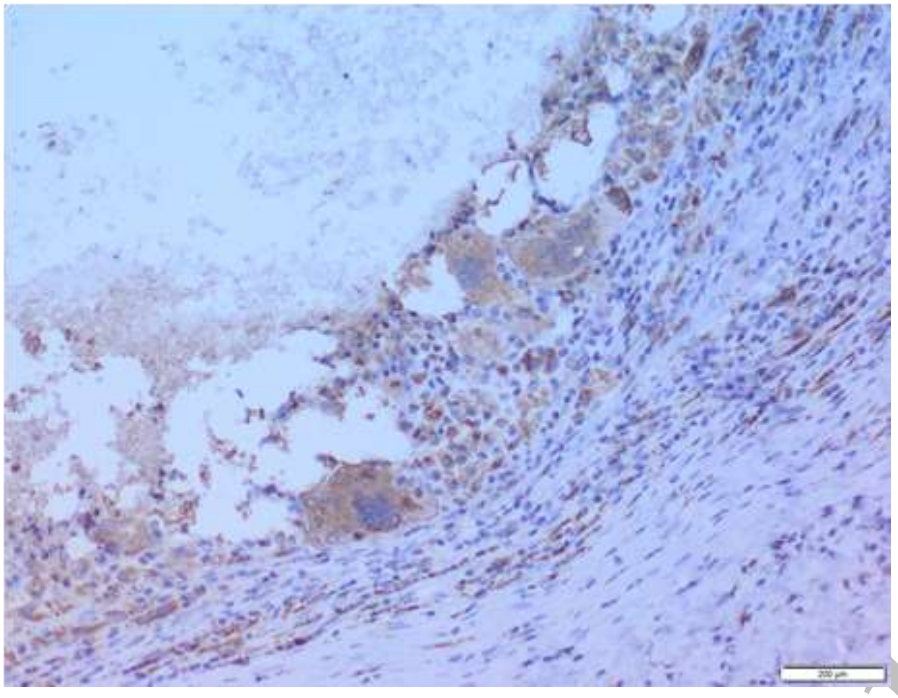

Figure 4. CD 68 positivity in multinucleated giant cells (immunohistochemistry,10×)

