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Exogenous chronic lipoid pneumonia: a case report

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Abstract

We present the case of a 75-year-old man who suffered from a post-stroke dysphagia and a long term history of exposure to paraffin oil for chronic constipation. At the chest computed tomography scan two pulmonary masses were detected, in the lower left lobe and in the middle lobe, respectively. Diffuse and bilateral ground-glass nodules were also described. A transbronchial biopsy and a bronchoalveolar lavage were performed. The histopathological examination showed the accumulation of lipoid material and macrophages with foamy and vacuolated cytoplasm surrounded by inflammatory cell infiltration and a mild fibrosis. The diagnosis of a exogenous chronic lipoid pneumonia was formulated. Reporting this case, we would like to alert the clinicians to be aware of this entity that can mimic other pathological conditions.

Key Words: lipoid pneumonia, lung disease, oils, lipid

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Introduction

Lipoid pneumonia is an uncommon form of pneumonia characterized by the presence of intra-alveolar lipid and lipid-laden macrophages on the bronchoalveolar (BAL) lavage, secondary to aspiration of various substances, such as petroleum jelly, mineral oils and nasal drops. The lack of specific clinical or radiological features may lead to missed or delayed diagnosis.

Case presentation

We report the case of a 75-year-old Caucasian man that complained of productive cough. The patient was a retired mechanic, former (quitted by 30 years) smoker of 15 p-y. A chest X-ray was performed, showing a lung opacity in the lower left lobe. An empiric wide spectrum antibiotic treatment was started for a suspected community-acquired pneumonia.

In his past history there was a diabetes mellitus (controlled with metformin and glibenclamide), systemic arterial hypertension (controlled with ramipril, hydrochlorothiazide, and nicardipine), chronic constipation with periodic use of paraffin oil and prostatic cancer, treated 10 years before with radical prostatectomy. An ischemic stroke also occurred 10 years before, with residual stroke-related disorders, dysphagia and multilacunar encephalopathy.

Following the appearance of persistent productive cough and exercise-induced dyspnoea, the patient presented at the Emergency Room of the Rovigo Hospital. A computed tomography (CT) scan of the chest showed the presence of a lung opacity in the lower left lobe and in the right middle lobe. A fiberoptic bronchoscopy and transbronchial biopsy of the lower right lobe were performed. The histopathological examination was negative for cancer cells, showing a non-specific inflammatory background. One month later the patient was referred to the Unit of Pulmonology of the University Hospital of Padova, Italy. At admission, the patient was asymptomatic. The laboratory examinations showed normal leukocyte count. The capillary blood gas test on room air was: pH 7.38, PaO2 69.2 mmHg, PaCO2 47.6 mmHg, SaO2 94%. Serum autoantibody panel was negative. Physical examination of the chest revealed bilateral and basal velcro-type crackles. No other pathological physical signs outside the chest were reported. Pulmonary function test results showed a restrictive pattern. A tree in bud pattern was detected at CT scan (Figure 1), mainly affecting middle and lower third of both lungs. At lower lobes, micronodules were tightly aggregate in patchy perihilar consolidations with air bronchograms, surrounded by ground glass, areas widely spreading along basal segments on the left lung. The consolidations showed a not homogeneously decreased attenuation with sampled values of -40/-50 Hounsfield Units (HU). Bronchiectasis, pleural effusion, mediastinal/hilar lymphadenopathy were not detected. The positron emission tomography (PET)-CT total-body highlighted an 8F-fluorodeoxyglucose (FDG) uptake in both lung masses, with standardized uptake values (SUV) of 9.65 in lower left lobe and 8.19 in the middle and lower lobe, respectively.

A fiberoptic bronchoscopy with endobronchial samples such as multiple trans-bronchial biopsies in pulmonary left lower lobe were performed. The bronchial aspirate fluid was sent for microbiological and mycobacteriological analysis that resulted negative. The biopsy specimen showed the accumulation of lipoid material, associated with inflammatory cell infiltration and a mild fibrosis, leading to the diagnosis of a lipoid pneumonia (Figure 2). A fairy amount of macrophages with foamy and vacuolated cytoplasm were detected in lung parenchyma, further supporting the diagnosis. Based on these results, an esophageal manometry was performed. The presence of hiatal hernia and hypotonia of the lower esophageal sphincter was reported. Moreover, the review of a CT scan of the

chest performed 4 years before the admission showed the same bilateral lung consolidations, thus favouring the diagnosis of a chronic disease.

Systemic glucocorticoid therapy, diet for dysphagia and the ban of oral laxatives were prescribed. The patient maintained stable clinical conditions for eighteen months. Following a rapid clinical worsening that started with gastrointestinal symptoms and fever, the patient was referred to another general hospital ward. The patient, after nineteen months, died for a cardiovascular acute event not related to lipoid pneumonia.

Fig.1 CT scan of the chest at the admission

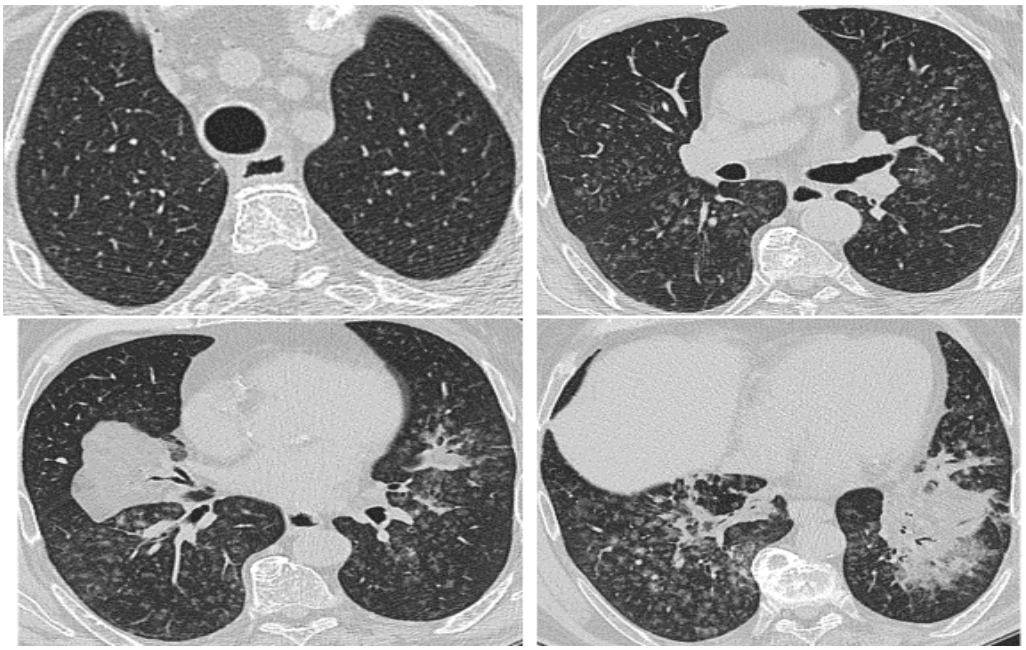
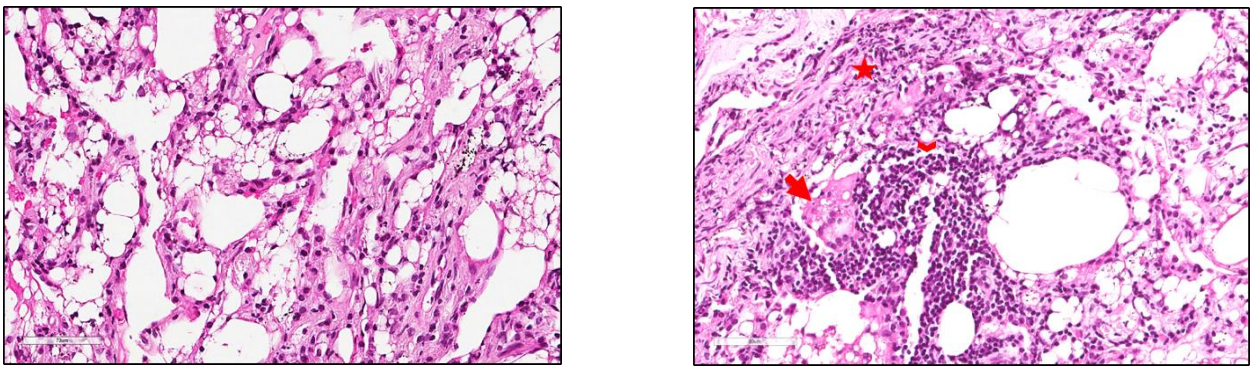


Fig.2 Histopathology of the transbronchial lung biopsy. Lung parenchyma showed the presence of lipoid material and macrophages with foamy and vacuolated cytoplasm, appearing as empty spaces (on the left, hematoxylin and eosin staining, 300 X original magnification). In adjacent areas, an inflammatory infiltrate and a mild fibrosis (star) were detected. Inflammatory background mainly consisted of lymphocytes (arrowhead) and some foreign-body giant cells containing cholesterol clefts (arrow) (Hematoxylin and eosin staining, 200 X original magnification).



Discussion

Lipoid pneumonia is a rare form of pneumonia characterized by the presence of intra-alveolar lipid. It is classified in an endogenous and exogenous form, according to the source of the lipids found in the lungs. Although uncommon, it is important to be aware of this entity to avoid misdiagnosis, as it can mimic other

pathological conditions (e.g. tumors). This problem mainly concerns the exogenous form. Indeed, the endogenous lipoid pneumonia is usually secondary to a known or suspected bronchial occlusion. Chronic exogenous lipoid pneumonia is often consequent to recurrent aspiration of oils (Table 1).

Tab.1 Most common causes and CT findings of chronic exogenous lipoid pneumonia

Most common causes of chronic exogenous lipoid pneumonia (Obtained with the data from 1, 5, 6, 13, 14)	
Laxatives Petroleum jelly Mouth sprays Oral lubricants Nose-drops Lip balm Ignition fluid (fire-eaters) Paraffine aerosol in iron foundry	**Mineral oils (obtained from petroleum)**
Milk Ghee (clarified butter) Cod liver oil Squalene (shark liver oil)	**Animal oils**
Vegetable glycerine (in e-cigarettes) Gingilli (sesame seed) oil Poppy seed oil Iodinated vegetable oil for bronchography Insecticide	**Vegetable oils**
Most common findings at CT scan of the chest of chronic exogenous lipoid pneumonia (Obtained with the data from 1, 2, 5, 15, 16)	
Single or multiple lung masses Single or multiple airspace consolidations Single or multiple ground-glass opacities Crazy-paving pattern Single or multiple aispace nodules	Lung consolidation with low density (-30 to-150 HU). Lobar distribution: middle lobe, right lower and left lower lobes, lingular lobe

Gastrointestinal alterations (such as diaphragmatic hiatal hernia), and neurological disorders with dysfunctional swallowing, can facilitate the aspiration (1).

Patients with chronic exogenous lipoid pneumonia are frequently asymptomatic and the disease is detected during chest imaging performed for other reasons.

Symptomatic patients frequently complain of cough or exercise-induced dyspnoea (2). Fever and weight loss are also reported (3). Physical examination of the chest is usually normal, but sometimes are present crackles (1).

The most common CT findings of the chronic exogenous lipoid pneumonia are summarized in Table 1.

Pulmonary consolidations have usually fat attenuation with low density (-30 to -150 HU) at CT scan of the chest. The main radiological differential diagnosis concern lung metastasis from chondrosarcomas and liposarcomas that can contain fat within the mass (2, 4, 5).

PET-CT usually showed an increased FDG uptake, highly suggestive of a lung malignancy (2,5,6,7).

Diagnosis of chronic exogenous lipoid pneumonia is often difficult because there are not specific clinical or radiological features. It is mainly based on the combination of history of exposure to oils, compatible radiological findings and the presence of lipid-laden macrophages in the sputum, in the BAL fluid and/or on lung biopsy (1, 5).

The radiologic findings of chronic lipoid pneumonia are usually slowly progressive (2).

Chronic lipoid pneumonia is often clinically indolent. In presence of concurrent debilitating illness and/or continued exposure to mineral oil, the disease may be progressive up to the formation of a mass (paraffinoma) or to a fibrotic process triggered by the inflammatory response. The disease can also be complicated by superinfection (5, 8).

The treatment of exogenous lipoid pneumonia is not well defined. It essentially consists of preventing further exposure to the mineral oil. The use of systemic glucocorticoid therapy has been recommended in some case reports (9, 10, 11, 12). Systemic glucocorticoid are initially administered at dose of 0.5 mg/Kg per day and continued for several weeks with gradual tapering (12). However, their use is limited to the patients with severe and progressive lung injury (1, 5, 8). The whole lung lavage is an alternative therapeutic option (12).

Conclusions

Exogenous lipoid pneumonia is a rare disease. Neverthless, it should be considered in patients with a long-term history of exposure to potential causative agents, particularly in patients with concomitant neurologic, in gastroesophageal and/or swallowing dysfunction, when lung masses with slow progression are detected.

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Conflicts of interest: The authors declare no conflict of interest.

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