Lipoid pneumonia with neuroendocrine tumorlet: a case report

Aldo Pezzuto,[1] Elisabetta Carico,[2] Alessia Pacini[1]

[1]Department of Cardiovascular and Respiratory Sciences, [2]Department of Clinical and Molecular Medicine, Sant'Andrea Hospital, Sapienza University of Rome, Italy

Abstract

Lipoid pneumonia is a rare disease. Herein we report an extremely rare case of lipoid pneumonia associated with a pulmonary tumorlet, located in the right lung which occurred in an 80-year-old man without any other organ involvement. The patient benefited from lung surgery and at a subsequent follow-up he had no consequences from surgery.

Correspondence: Aldo Pezzuto, Department of Cardiovascular and Respiratory Sciences Sant'Andrea Hospital, Sapienza University of Rome, via Grottarossa 1035, Rome, Italy.
Tel. +39.06.6640052. E-mail: aldo.pezzuto@uniroma1.it

Key words: lipoid pneumonia, pulmonary tumorlet, thoracic surgery.

Contributions: all the authors made a substantive intellectual contribution. All the authors have read and approved the final version of the manuscript and agreed to be accountable for all aspects of the work.

Conflict of interest: the authors declare no potential conflict of interest.

Funding: none.

Availability of data and materials: all data generated or analyzed during this study are included in this published article.

Ethics approval and consent to participate: written informed consent was obtained from the patient for the publication of this manuscript and any accompanying images.

Acknowledgments: we express our gratitude toward colleagues radiologist and pathologist.

Received: 11 August 2023.
Accepted: 23 August 2023.
Early view: 27 August 2023.

Licensee PAGEPress, Italy
Chest Disease Reports 2023; 11:11653
doi:10.4081/cdr.2023.11653

Introduction

Lipoid pneumonia is a relatively rare disease, characterized by the accumulation of adipose material at the alveolar and interstitial levels. The pathogenesis is linked to the aspiration or inhalation, chronic or occasional, of oily substances. It can present with symptoms such as cough, fever, breathlessness, chest pain, but it can sometimes be asymptomatic.[1,2] It is important to exclude a lung cancer.[2]

Case Report

An 80-year-old man, former smoker and retired chemist, presented himself at our attention complaining of chest tightness in the last two months. His chest Computed Tomography (CT) scan showed an expansive solid formation with irregular morphology, located in the lower right lobe in peri-hilar site. It was characterized by non-homogeneous values of density, due to the presence of solid tissue mixed with fluid density areas that were likely necrotic, with a heterogeneous enhancement after contrast medium administration. Ground glass opacities, bronchiectasis, lymphadenopathies in pre and para- tracheal and subcarinal station were found, too. A necrotic substance appeared inside (Figure 1 A-C). After a few days, the patient underwent a bronchoscopy with transbronchial biopsies in the right lower lobe. The microbiological examination showed no pathogenic flora and negative research for mycobacteria as well. The histological examination showed fragments of bronchial mucosa and lung parenchyma free from neoplastic infiltration, but the site of lipoid pneumonia. The cytological preparations consist of upper respiratory tract cells, neutrophil granulocytes, macrophages lung and some normo-typical bronchial elements. After thoracic surgeon evaluation, since there was a suspected neoplastic pathology, the patient performed a spirometry that revealed normal values and subsequently he underwent a lower right lobectomy. The histological examination showed: lung parenchyma characterized by homogeneous fibrosis, with thickening of alveolar septa that sometimes show "swiss cheese-like" appearance, lymphoplasmacytic phlogosis at interstitial arrangement, accumulation of macrophage foamos in endoalveolar and interstitial locations. The morphological picture, also in the light of the clinical suspicion, is compatible with a phlogistic process in the type of lipoid pneumonia. Absence of definite images referrable to neoplasm in the material under examination. Present, in the surrounding lung parenchyma, neuroendocrine tumorlet (CD56+, Sinatophysin+, Chromogranin+) with a diameter of 0.2 cm. Station 4, 7, 9 and 10 lymph nodes sent separately free of images referrable to neoplasia. Laboratory findings were notable for a markedly elevated serum C reactive protein (10 mg/dL), an increased level of white blood cells (12,000/mm^3), Covid-19 swab test was negative. After three months, the patient's clinical condition was good except for mild dyspnea on exertion, even though no respiratory failure was developed.

Discussion

Lipoid pneumonia results from the accumulation within alveolar macrophages of endogenous or exogenous lipids.[3]

The diagnosis of lipoid pneumonia is based on typical radiological aspects, with particular attention to the study of density, and an adequate clinical history correlation. Radiological appearance of the disorder can resemble many other lung diseases; therefore, histological confirmation is sometimes necessary to rule out a malignant origin.[3,4]

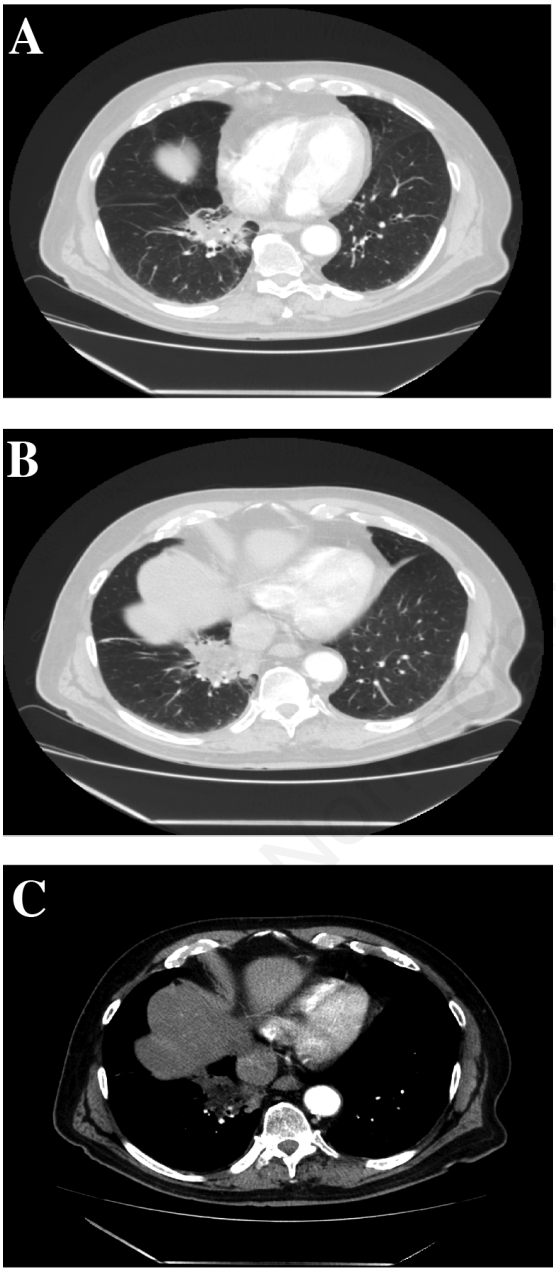

Figure 1. A) Computed Tomography (CT) scan revealing an opacity located in inferior right lobe. **B**) CT scan revealing in relation with descending aorta. **C**) CT scan revealing a necrotic substance inside.

The clinical symptoms could be various, inducing to seek a differential diagnosis. Sometimes it is asymptomatic, needing a clinical history evaluation to find the causes of lipid inhalation. The reported case shows how inhalation of foreign substances can generate a strong chronic inflammatory reaction, bronchiectasis and fibrosis. Since the patient underwent surgical resection and a neuroendocrine tumorlet was found, it would be suggestive to promote new studies investigating the causes of a possible association of lipoid pneumonia and tumorlet, which is a pulmonary neuroendocrine cell hyperplasia. The latter could foster the development of a neuroendocrine tumor, such as carcinoid. Its finding is probably due to the strong chronic inflammation induced by the lipid material deposition and it can be found in association with infectious diseases such as tuberculosis or with other carcinoids.[5,6] Differential diagnoses should be kept in mind with several other forms of lung thickening or nodule, such as those associated with tobacco smoking or those associated with pulmonary fibrosis.[7] We know that respiratory complications could be found due to surgery, however in our case report no respiratory failure was observed three months after lung surgery, since the patient had a normal respiratory function before surgery.[8]

Conclusions

When evaluating a lung opacity that presents with chest pain, we must place the differential diagnosis with pneumonia and lung cancer. The association of a tumorlet requires a specific surgical approach.

References

1. Kono K, Hamaguchi M, Tanino A, et al. Lipoid pneumonia caused by esophageal achalasia. Intern Med 2023. Online ahead of print.
2. Costa FMD, Cerezoli MT, Medeiros AK, et al. Small samples, big problems: lipoid pneumonia mimicking lung adenocarcinoma. J Bras Pneumol 2023;49:e20230147.
3. Marchiori E, Zanetti G, Mano CM, Hochhegger B. Exogenous lipoid pneumonia. Clinical and radiological manifestations. Respir Med 2011;105:659-66.
4. Betancourt SL, Martinez-Jimenez S, Rossi SE, et al. Lipoid pneumonia: spectrum of clinical and radiologic manifestations. AJR Am J Roentgenol 2010;194:103-9.
5. Onda T, Kita H, Yoshida T, Hayashi T. Pulmonary tumorlet associated with atypical Mycobacterium. Kyobu Geka 2023;76:432-5.
6. Maki Y, Okada K, Nakamura R, et al. A case of multiple lung carcinoid tumors localized in the right lower lobe. Respir Med Case Rep 2022;38:101679.
7. Pezzuto A, Tammaro A, Salerno G, et al. Lung nodule and pulmonary fibrosis at first evaluation in smoker undergoing a smoking cessation program. Global J Respir Care 2022;8:11-3.
8. Pezzuto A, Trabalza Marinucci B, Ricci A, et al. Predictors of respiratory failure after thoracic surgery: a retrospective cohort study with comparison between lobar and sub-lobar resection. J Int Med Res 2022;50:3000605221094531.