Exogenous Lipoid Pneumonia

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Abstract

Background: Lipoid pneumonia is a rare disease and is usually reported as sporadic cases. *Exogenous lipoid pneumonia* is a more common form of lipoid pneumonia. Untypical characteristics make incidence rate of these cases often imprecise. The diagnosis is made from a history of lipid-containing material aspiration, radiologic imaging, and histopathological examination. There is no treatment of choice because diagnoses of these cases are rarely made.

Case: We report one case at Dr. Saiful Anwar hospital, a 35 years old man with accidental kerosene ingestion and aspiration. Based on examination of the patient, laboratory results, radiological imaging, and BAL fluid analysis and cytology, a diagnosis of exogenous lipoid pneumonia was made. Our treatment of choice was a combination of antibiotics, corticosteroids and BAL to manage this patient. After 2 weeks of treatment, there were improvements in clinical and radiological imaging.

Conclusion: Exogenous lipoid pneumonia is a rare disease. Using antibiotics, corticosteroids, and BAL through bronchoscopy can be a therapeutic option that provides clinical and radiological improvement.

Keywords: Lipoid Pneumonia, Exogenous Lipoid Pneumonia, Treatment of Lipoid Pneumonia.

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1. Introduction

Kerosene is one of the hydrocarbon compounds that was widely used in 19th century as household fuel, but getting less and less with the development of electric and gas power[1]. Like other chemical compounds, exposure to kerosene able to cause health effects at a certain threshold. Aspiration of its liquid form can cause damage to the lung, and cause pneumonitis known as exogenous lipoid pneumonia[2]. Exogenous lipoid pneumonia is a rare disease, found as sporadic cases[3]. Diagnosis for these cases is usually challenging, especially in a rural area. Here, we report a rare case of exogenous lipoid pneumonia caused by kerosene aspiration.

2. Case Report

A 34-year-old man was referred to our hospital with a chief complaint of

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bloody cough. He had been suffering from bloody cough since 5 days before admission, after unintentionally drinking approximately 30 ml of liquid kerosene. He also felt shortness of breath 2 days later, followed by chest pain in the right hemithorax without any referred pain. He is an ex-smoker, stop smoking 2 years before, and never had any respiratory diseases before. He took self-medication with an antitussive agent and antacid but the symptoms did not resolve. His saturation was 98% without oxygen supplementation, chest examinations were within normal limit without any abnormal lung sounds.

Table 1. Laboratory Data on admission

	Values
Hematology	
Leucocyte (cells/ uL)	13,560
Hemoglobin (g/dL)	13.1
Hematocrit (%)	37.8
Platelet (cells/ uL)	189,000
MCV (fL)	87.7
MCH (Pg)	30.4
MCHC (g%)	34.7
Eosinophil (%)	0.1
Basophil (%)	0.1
Neutrophil (%)	80.9
Lymphocyte (%)	11.9
Monocyte (%)	7
Biochemistry Tests	
Random blood sugar	105
(mg/dL)	23.7
Ureum (mg/dL)	0.68
Creatinine (mg/dL)	15
SGOT (U/L)	10
SGPT (U/L)	0.32
Total bilirubin	0.09
(mg/dL)	0.23
Direct bilirubin	3.53
(mg/dL)	133
Indirect bilirubin	4.13
(mg/dL)	110
Albumin (g/dL)	4.2
Sodium (mmol/L)	
Potassium (mmol/L)	
Chloride (mmol/L)	
Procalcitonin (ng/ml)	
Arterial Blood Gas	
Analysis (fiO2 28%)	7.34
pH	32
PCO2 (mmHg)	113
PO2 (mmHg)	17.6
HCO3- (mmol/L)	-8.3
BE (mmol/L)	98.7
Sat O2 (%)	

His leucocyte count was increase (13,500 cells/uL) with elevated neutrophil percentage (80.9 %). His renal function and liver function test were within normal limit, procalcitonin level was increase (4.2 ng/mL). Blood gas analysis with 28% oxygen fraction shows slightly acidemia (pH : 7.34), slightly decrease of pCO2 (32 mmHg), increase of PO2 (113 mmol/L), a decrease of HCO3 and base excess (respectively 17.6 mmol/L and -8.3 mmol/L). Lung function was measured and revealed normal results, as shown in figure 1.

Table 2. Analysis of bronchoalveolar lavage's fluid.

Component	Values
Total Protein (g/dL)	1.04
Glucose (mg/dL)	7
LDH (IU/L)	519
Macroscopic	
Color	Pink
Clot	Positive
Clarity	Slightly cloudy
Microscopic	
Erythrocyte (cells/ uL)	4,850
Leucocyte (cells/ uL)	20
PMN (%)	0
MN (%)	100

Radiographic examination are taken at the admission and shows infiltrate in the middle and lower part of left hemithorax. High resolution computed tomography of

lung revealed consolidation at segment 6 of right lung with surrounding ground glass opacity. Fiber optic bronchoscopy was done in the second day after admission. There were erythematous macules in almost all bronchial mucosa especially in the truncus intermedius of the right lung. Segmental bronchoalveolar lavage (BAL) was done with normal saline and cytologic sample was taken. Analysis of BAL's fluid shown in table 2. Sample taken during bronchoscopy also checked for microbiology examination and later, the results shown no acid-fast bacillus, negative rapid molecular testing for tuberculosis (geneXpert), and no growth of bacterial in the culture. Cytology examination revealed lipid laden macrophage and the diagnosis of exogenous lipoid pneumonia was confirmed.

Figure 1. Body plethysmograph shows normal lung function

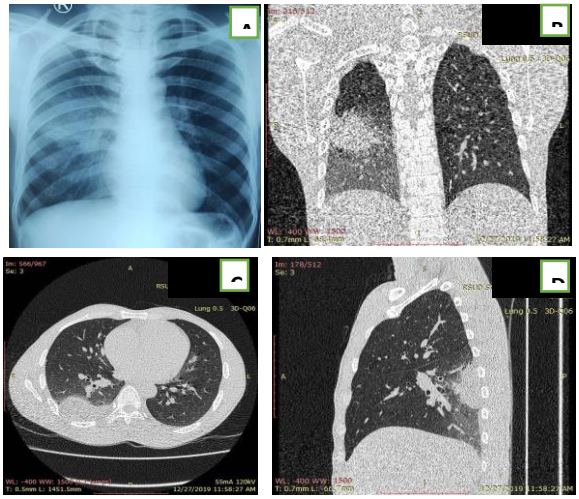

Figure 3. A : Chest radiography at admission shown infiltrate in the middle and lower fields of the right lung. B,C,D : High-Resolution Chest Computed-tomography revealed consolidation at supero-posterior segment, inferior lobe of right lung.

Beside segmental BAL with normal saline, an antibiotic (injection of Ceftriaxone 2 x 1 gram) and steroid (injection of methylprednisolone 2 x 62.5 mg) were administered for 5 days. Supplemental therapy such as oxygenation and symptomatic treatments were also given. Five days after admission, the clinical condition was improve and patient can be treated on an outpatient basis. He didn't have any symptoms 2 weeks later, and chest radiology revealed improvement as shown in figure 4.

3. Discussion

Exogenous lipoid pneumonia is a rare form of pneumonia, cause by inhalation of fatty substances. The clinical incidence for this cases are unknown, but estimated 1-2.5

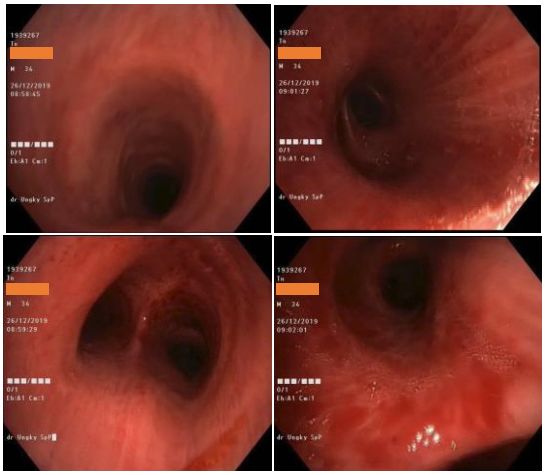

Figure 2. Bronchoscopy shown erythematous macules in bronchial mucosa

% based on post-mortem study[3]. Risk factor of exogenous lipoid pneumonia are younger children, have a local anatomical defect such as palate cleft, functional impairment of faring, neuromuscular abnormality causing swallow or cough disorder, fire breathers, or routinely using lipid containing substances such as lip balm, petroleum jelly, and oil-based laxative.[4,5] Exogenous lipid pneumonia can be classified furthermore as an acute or chronic form. Chronic pneumonia are usually caused by routinely aspirate fatty substances in small quantities (or micro-aspiration) but for a long period of time.[5] Otherwise, acute form is usually come in the event of an accidental aspiration of large quantities fatty substances[6]. Pathologically, lipid pneumonia occurred as an inflammatory reaction to foreign materials[5]. The parenchymal damage in lipid pneumonia depends on type, quantities, fre-

quency, and duration of aspiration or inhalation of fatty or oily substances. Vegetable and mineral oil are usually cause minimally inflammation. But, animal fats are usually causing severe inflammation reaction, manifested as focal edema with intra-alveolar bleeding[3]. Aspiration or inhalation of fatty substances didn't induce cough reflex and cause disruption of muco-ciliary system. When it enters the alveoli, lipid material are phagocyted by macrophage after emulsified process. Macrophage itself are unable to metabolized lipid, and after apoptosis of macrophage cell, lipid are released back into the alveoli. This repeated process induce inflammation, causing parenchymal destruction, and later, turn into fibrosis[5].

Diagnosis of exogenous lipid pneumonia based on its clinical presentation, cyto-pathology examination from bronchoscopy sample, radiographic patterns and history of aspiration or inhalation fatty substances. In chronic exogenous lipoid pneumonia, the symptoms are usually insidious. The clinical presentation in acute disease is various, ranging from mild symptoms until severe conditions required mechanical ventilation.[3,5] Beside respiratory symptoms, other extra-thoracal symptoms such as nausea, vomiting, abdominal pain, dizziness, fever (usually low grade), and syncope are reported in several literatures.[6,11] Radiographic abnormalities, using chest CT are found in 96% of cases, based on one study, which 86% of cases is consolidation, 47% is ground glass appearance, 22% is intra-alveolar nodule, and 20% is crazy paving pattern.[8] This feature resembles many other lung diseases such as lung tuberculosis, malignancy, pneumonia, or acute respiratory distress syndrome. The diagnosis accuracy is lower when only using chest x-ray as radiographic approach.[5,6] High-resolution computed tomography is the most desirable radiography modality to diagnose lipoid pneumonia[9]. Lower lobe of right lung are the most common location related to exogenous lipoid pneumonia because its large caliber and more vertical in position[10].

Bronchoscopy are important tools for diagnosis exogenous lipoid pneumonia. Not only for diagnostic approaches, it can be used for management by broncho-alveolar lavage. Erythematous and edema mucosal are most common abnormalities found, especially in middle and lower lobe. Several studies report

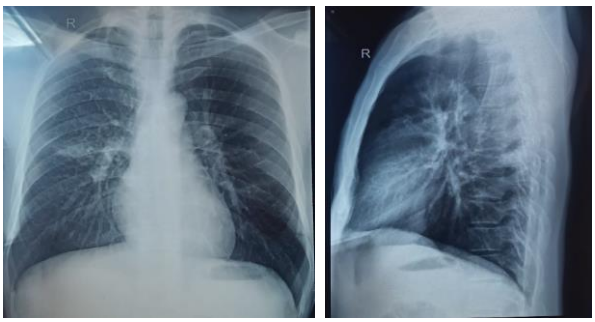

Figure 4. Chest radiography taken 2 weeks after treatment revealed reduction of infiltrate in right lung

a whitish-grey secretion which describes oily material in airway.[11] Sample obtained from bronchoscopy or BAL procedure are usually muddy whitish and appears to have turned into two layers with lump of fat at top[12]. Furthermore, cytology examination for this sample revealed lipid laden macrophage, that able to confirm the diagnosis of lipoid pneumonia. Beside BAL, sputum of patients can also be used to search for lipid laden macrophage.[4,11] Identification of lipid contained in lipid laden macrophage is clearer when using lipid stain such as oil red o or black sudan staining[13].

Our cases are diagnose with exogenous lipoid pneumonia based on history of aspiration of kerosene, which is oily substances, the consolidation and ground glass pattern in lower lobe of right lung based on radiographic examination, and presence of lipid laden macrophage from BAL sample. Even though we didn't find any whitish-grey secretion or oily substances during bronchoscopy, but other findings are enough to diagnose our cases with exogenous lipoid pneumonia.

Our cases are treated with supplemental therapy such as oxygenation and symptomatic treatment, combine with antibiotic and steroid administration, also with segmental lavage. In many cases reported, supplemental therapy is a main modality to treat mild exogenous lipoid pneumonia[14]. But several case also felt in serious condition that need mechanical ventilation to support respiratory system. Corticosteroid is a main choice for anti-inflammatory and usually show promising results. However, it is not recommended to be given in all cases of lipoid pneumonia. Patient with mild respiratory symptoms usually not required to get corticosteroid. In several studies, corticosteroid administration didn't have any impact on clinical condition or radiologic improvement.[11,14,15] Dosage and duration of corticosteroid are not detailed in many cases. But some literatures recommend to give 0.5 mg/ kg body weight/ day with gradual dose reduction. The duration are variable from several day up to six months, depend on clinical and radiographic improvement of the patient.[11,16] Corticosteroids are not recommended to be given in cases where fibrosis is the main or only feature on radiology examination.[8]

Antibiotics administration are usually given in severe cases because its frequently complicate with bacterial infection, or in any other condition where a bacterial pneumonia is also suspected.[16] Previous study recommend to give penicillin as prophylaxis in lipoid pneumonia, but latest report didn't recommend to give antibiotic as prophylaxis measures of bacterial infection.[11,16,17]

The main purpose of doing broncho-alveolar lavage is to clear the airway from fatty substances. Whether done as segmental or whole lung lavage, both give good results on several reports.[18,19] Improvement that occurred after segmental lavage explained that BAL can improve the conditions not only by cleaning the airway, but also through an unexplained mechanism.[18] The choice of solution used in BAL was also investigated in several studies, but normal saline was considered to be the most effective in most cases.[15]

4. Conclusion

In conclusion, our case is diagnose with exogenous lipoid pneumonia based on history of aspiration lipid-containing material, presentation of radiographic abnormalities, and presence of lipid laden macrophage from BAL sample. The combination of these three (history taking, radiology features, and presence of lipid laden macrophage) should be considered as the standard in establishing the diagnosis of lipoid pneumonia. We also recommend a combination of steroids, supplemental therapy, and BAL as the management of exogenous lipoid pneumonia, whereas antibiotics may considered if there are any signs of infection (in this case, clinical sign, radiologic features, and elevated of leukocytes and procalcitonin are the consideration for the administration of antibiotics).

References

1. Lam, N. L., Smith, K. R., Gauthier, A., & Bates, M. N. (2012). Kerosene: A review of household uses and their hazards in low-and middle-income countries. Journal of Toxicology and Environmental Health - Part B: Critical Reviews, 15(6), 396–432.
2. Chilcott, R. P. (2006). Health Protection Agency Compendium of Chemical Hazards: Kerosene (Fuel Oil). Public Health England, 1–31.
3. Betancourt, S. L., Martinez-Jimenez, S., Rossi, S. E., Truong, M. T., Carrillo, J., & Erasmus, J. J. (2010). Lipoid pneumonia: Spectrum of clinical and radiologic manifestations. American Journal of Roentgenology, 194(1), 103–109.
4. Sung, S., Tazelaar, H., Crapanzano, J., Nassar, A., & Saqi, A. (2018). Adult exogenous lipoid pneumonia: A rare and underrecognized entity in cytology - A case series. CytoJournal, 15(1).
5. Hadda, V., & Khilnani, G. C. (2010). Lipoid pneumonia: An overview. Expert Review of Respiratory Medicine, 4(6), 799–807.
6. Marchiori, E., Zanetti, G., Mano, C. M., & Hochhegger, B. (2011). Exogenous lipoid pneumonia. Clinical and radiological manifestations. Respiratory Medicine, 105(5), 659–666.
7. Kim, C. H., Kim, E. J., Lim, J. K., Yoo, S. S., Lee, S. Y., Cha, S. I., … Lee, J. (2018). Comparison of exogenous and endogenous lipoid pneumonia: The relevance to bronchial anthracofibrosis. Journal of Thoracic Disease, 10(4), 2461–2466.
8. Beck LR, Landsberg D. Lipoid Pneumonia. [Updated 2020 Jun 22]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2020 Jan-.
9. Doubková, M., Doubek, M., Moulis, M., & Skřičková, J. (2013). Exogenous lipoid pneumonia caused by chronic improper use of baby body oil in adult patient. Revista Portuguesa de Pneumologia, 19(5), 233–236.
10. Gamache, J., Kamangar, N., Hoo, G., Varkey, B., Grier, L., Stearns, D., Varkey, A. and Swaminathan, A., 2018. Aspiration Pneumonitis And Pneumonia: Overview Of

Aspiration Pneumonia, Predisposing Conditions For Aspiration Pneumonia, Pathophysiology Of Aspiration Pneumonia. [online] Emedicine.medscape.com. Available at: <https://emedicine.medscape.com/article/296198-overview#a1>

11. Kim, H., Lee, W. H., Jeong, N., Lee, J. H., & Park, J. H. (2018). Lipoid Pneumonitis in a patient with an accidental ingestion of kerosene successfully treated with bronchoscopic segmental lavage and systemic steroid.
12. Kuroyama, M., Kagawa, H., Kitada, S., Maekura, R., Mori, M., & Hirano, H. (2015). Exogenous lipoid pneumonia caused by repeated sesame oil pulling: A report of two cases. BMC Pulmonary Medicine, 15(1), 1–5.
13. Gotanda, H., Kameyama, Y., Yamaguchi, Y., Ishii, M., Hanaoka, Y., Yamamoto, H., … Ouchi, Y. (2013). Acute exogenous lipoid pneumonia caused by accidental kerosene ingestion in an elderly patient with dementia: A case report. Geriatrics and Gerontology International, 13(1), 222–225.
14. Yasui, H., Yokomura, K., & Suda, T. (2016). A severe case of acute exogenous lipoid pneumonia treated with systemic corticosteroid. Respiratory Medicine Case Reports, 17, 64–67.
15. Russo, R., Chiumello, D., Cassani, G., Maiocchi, G., & Gattinoni, L. (2006). Case of exogenous lipoid pneumonia: Steroid therapy and lung lavage with an emulsifier. Anesthesiology, 104(1), 197–198.
16. Guo, M., Liu, J., & Jiang, B. (2019). Exogenous lipid pneumonia in old people caused by aspiration: Two case reports and literature review. Respiratory Medicine Case Reports, 27(April), 100850.
17. Gentina T, Tillie-Leblond I, Birolleau S et al. 2001 Fire-eater's lung: seventeen cases and a review of the literature. Medicine (Baltimore), 80, 291–297.
18. Nakashima, S., Ishimatsu, Y., Hara, S., Kitaichi, M., & Kohno, S. (2015). Exogenous lipoid pneumonia successfully treated with bronchoscopic segmental lavage therapy. Respiratory Care, 60(1), e1–e5.
19. Lu, G., Xie, Y., Huang, L., Tong, Z., Xie, Z., Yu, J., & Wu, P. (2016). Study of Acute Exogenous Lipoid Pneumonia. Indian Journal of Pediatrics, 83(8), 787–791.