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Case report

A severe case of acute exogenous lipoid pneumonia treated with systemic corticosteroid

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ABSTRACT

Acute exogenous lipoid pneumonia is a rare disorder in adults. A treatment of choice for lipoid pneumonia has not been established, and systemic corticosteroid use remains controversial. We report the case of a 32-year-old man with schizophrenia who presented with kerosene-induced acute exogenous lipoid pneumonia that was treated with a systemic corticosteroid. In this case, supportive therapy did not improve the patient's condition, so systemic corticosteroid therapy was commenced four days after he ingested the kerosene. After corticosteroid commencement, the patient's symptoms and hypoxia improved within a few days. Although some radiological characteristics of this disorder have been reported previously, the process of radiological improvement of exogenous lipoid pneumonia is not well known. In this case, computed tomography findings changed dramatically after corticosteroid therapy was initiated. Extensive bilateral consolidations that were observed on admission improved. Although pneumatoceles developed two weeks after corticosteroid commencement, they were nearly gone after two months of the treatment. While corticosteroid therapy is not suitable for all cases, it should be considered for severe or refractory cases.

1. Introduction

Acute exogenous lipoid pneumonia is a rare disorder caused by aspiration of lipid formulations, mostly attributed to accidental ingestion. Clinical courses of acute exogenous lipoid pneumonia range from asymptomatic to life threatening, based on the amount and quality of oil aspirated [1]. Although some radiological characteristics of exogenous lipoid pneumonia have been reported in the literature [2,3], little is known about the process of radiological improvement. There is no consensus regarding the choice of treatment for this disorder; treatment is primarily supportive and generally conservative. Although systemic corticosteroid use remains controversial [4], treatment with a corticosteroid may lead to rapid improvement in some active cases [5,6].

This report describes a severe and refractory case of acute exogenous lipoid pneumonia following kerosene aspiration, which was treated with a systemic corticosteroid. The patient's symptoms improved within a few days of starting corticosteroid treatment. The radiological abnormalities observed were dynamically changed and nearly resolved two months after treatment initiation.

2. Case report

A 32-year-old man ingested kerosene (approximately 100 mL) from a plastic bottle. Coughing and choking thereafter resulted in kerosene aspiration. The patient also suffered nausea and vomiting after ingesting kerosene. Over time, chest pain and chills emerged and worsened. Fifteen hours after ingestion, the patient was brought to the emergency room. The man had a history of schizophrenia and was taking some medications. On physical examination, the pulse was recorded as 108 beats/min, with a respiratory rate of 30 breaths/min and body temperature of 40.0 °C. Auscultation revealed diminished respiratory audibility. The patient's breath and stool smelled of kerosene. Arterial blood gas analysis revealed type I respiratory failure; therefore, providing 5 L/min of oxygen via a face mask was necessary. SpO_2 was 93% under this condition. Laboratory findings showed a lactate dehydrogenase level of 365 IU/L and a C-reactive protein level of 14.1 mg/dL. A

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chest radiography revealed bilateral infiltrations (Fig. 1), and a computed tomography (CT) scan showed bilateral airspace consolidations, predominantly in the right lower lobe (Fig. 2). Extracellular oily droplets were found on a sputum cytological examination (Fig. 3). Blood and sputum cultures were negative. Therefore, we diagnosed exogenous lipoid pneumonia based on the patient's clinical history as well as the radiological and cytological findings. Despite supportive therapy with a broad-spectrum antibiotic, the symptoms and hypoxia failed to improve. Based on empirical evidence, 30 mg prednisolone therapy was commenced four days after kerosene ingestion. Within a few days, the symptoms and hypoxia were improved. Two weeks after the initiation of prednisolone treatment, the consolidations seen on the chest CT scan were improved, although some cavity lesions (pneumatoceles) developed in the right lung (Fig. 4A). Prednisolone was tapered to every other week, and was completed two months after initiation. Two months after the corticosteroid therapy commencement, the consolidations and pneumatoceles had disappeared, and only pleural and interlobular septal thickening remained (Fig. 4B). No recurrence has been observed in the patient so far.

3. Discussion

Exogenous lipoid pneumonia is an uncommon pulmonary disorder that results from the accumulation of lipids in the alveoli [3,7]. Aspiration of a large quantity of lipid material during a short period of time results in acute exogenous lipoid pneumonia, while long-term recurrent inhalation exposure to oil products results in the chronic form [8]. Acute exogenous lipoid pneumonia is mostly attributed to accidental ingestion by children [3]. Of the children who ingest hydrocarbons, approximately 40% develop significant pulmonary signs and symptoms [9]. Kerosene is well known to cause pulmonary complications, and is a representative hydrocarbon because it is incorporated into many common household products and is volatile [9]. Kerosene is a low-viscosity hydrocarbon liquid that spreads over large areas of the lining of the lungs. Aspiration of kerosene primarily induces inflammation and loss of surfactant [10]. The death rate for kerosene aspiration pneumonia is as high as 4%–10% [11]. In adults, near-drowning incidents in a kerosene-contaminated river [12], a suicidal attempt [13], and an accidental ingestion by an elderly patient with dementia [14] have been reported.

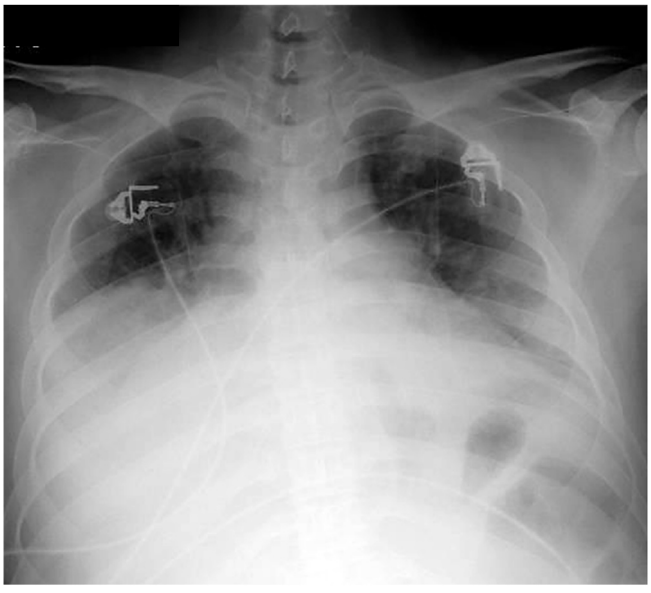

Fig. 1. Chest radiograph on admission shows bilateral infiltrations, predominantly in the lower lung field.

A diagnosis of lipoid pneumonia is based on a history of exposure to oil with radiological findings consistent with the disease and the presence of lipid-laden macrophages on sputum or bronchoalveolar lavage (BAL) analysis [15,16]. In the patient described in this report, a history of kerosene ingestion was obvious. Both his breath and stool smelled of kerosene. Lipid-laden macrophages were not apparent in his sputum, but extracellular oily droplets were observed. The reason was thought to be the timing of the sputum sampling; the sputum was collected within 24 h of kerosene ingestion in this patient. Marchiori et al. have shown that the presence of extracellular oily droplets is more specific for exogenous lipoid pneumonia [8]. The most characteristic CT finding of exogenous lipoid pneumonia is the presence of consolidations with low attenuation [2]. In this patient, the extensive pulmonary consolidations were detected in the lower lobes and the right middle lobe on admission. However, low attenuation in the consolidations was not obvious. The reason was thought to be intense inflammation occurring in the lung parenchyma. Densities of the consolidations were, therefore, higher compared to previous reports. Consolidations were improved two weeks after corticosteroid treatment initiation. On the other hand, pneumatoceles emerged in the right lung accompanied by niveau formation seen on the CT scan. Similarly, the development of pneumothorax, pneumatoceles, and bronchopleural fistulas has been reported as secondary effects of kerosene aspiration in some cases [10,13]. Two months after the initiation of corticosteroid treatment, the consolidations and pneumatoceles disappeared, and only some bronchiectasis remained, with pleural and interlobular septal thickening seen on the CT scan.

A treatment of choice for lipoid pneumonia is not well established. Treatment recommendations are based on clinical experience rather than long-term observational studies. BAL has therapeutic implications in some cases [17]; however, this patient had severe respiratory failure, making therapeutic BAL to remove the agent difficult. Moreover, the role of therapeutic BAL in acute cases has yet to be evaluated. Although corticosteroid therapy has been used successfully in some cases [5,6,18], in canine studies, corticosteroid therapy did not have an effect on kerosene aspiration [9]. Since this patient had schizophrenia treated with antipsychotic drugs, corticosteroid therapy was avoided when he was admitted. However, the symptoms and hypoxia did not respond to supportive therapy. His respiratory status worsened and he was exhausted due to dyspnea and fever. Pulmonary damage reaches its clinical peak approximately 3 days after the hydrocarbon aspiration [19]. In this case, lung injury was severe and continued to worsen after this period; therefore, systemic corticosteroid treatment was commenced four days after kerosene ingestion under the observation of a psychiatrist. Dosing and duration of the corticosteroid for lipoid pneumonia are not precisely established. We commenced a 30 mg per day (60 kg man at a dose of 0.5 mg/kg) prednisolone treatment based on previous reports [5,6]. Corticosteroid therapy resulted in rapid improvement and fever lysis in this patient, and CT findings were improved after two months of corticosteroid treatment. In this case, the duration of corticosteroid therapy (two months) was short compared to previous reports [5,6], but recurrence was not observed after the treatment was completed.

Although the value of corticosteroid therapy for exogenous lipoid pneumonia has never been clearly established, there are reports of the use of systemic corticosteroids to slow the inflammatory response in diffuse and severe cases [6,18]. Hussain et al. reported that radiography and lung function were recovered normally in a patient with severe exogenous lipoid pneumonia

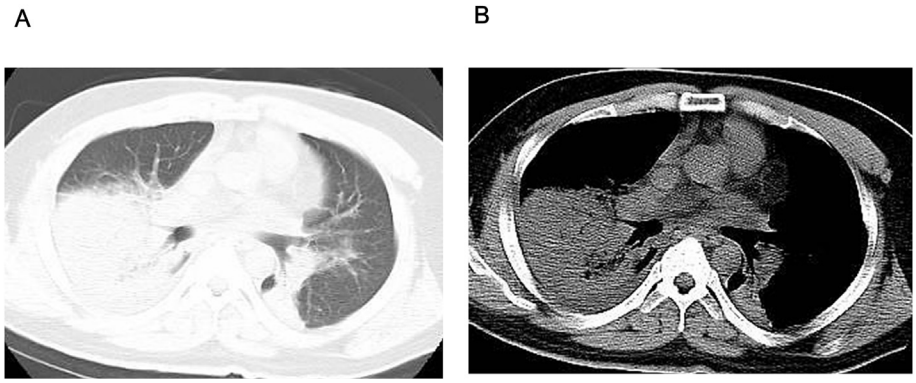

Fig. 2. Chest computed tomography scan on admission reveals airspace consolidations with air bronchograms in the lower lobes and right middle lobes (lung window) (A). These consolidations are of the same densities as those of the muscle and heart in the mediastinal window (B).

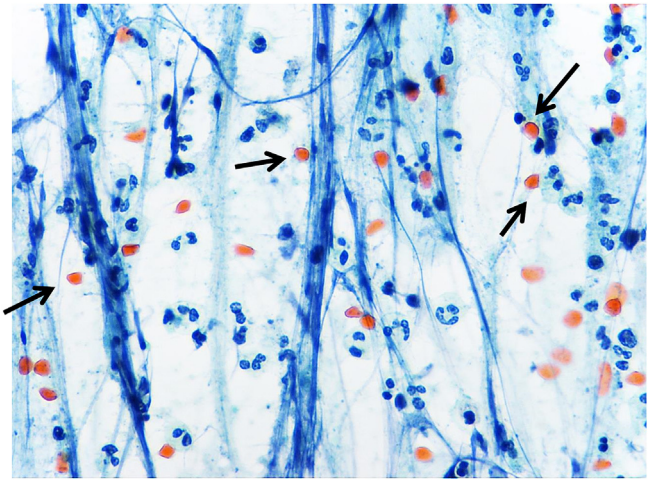

Fig. 3. Sputum cytology shows extracellular oily droplets (arrows) (Oil Red O Stain: red is positive).

There is the possibility that corticosteroid therapy prevents fibrosis and loss of lung volume in acute severe cases. On the other hand, corticosteroid may not be effective once the disease has progressed to fibrosis. In fact, corticosteroid therapy induced only a slight improvement in a case with repeated sesame oil pulling [20]. In another case, prednisolone did not improve symptoms and pulmonary function in a patient applying petroleum jelly intranasally for 5 years [21].

Corticosteroid therapy for exogenous lipoid pneumonia has not been effective in all reported cases. In fact, systemic corticosteroid therapy may bring several unfavorable side effects, especially increased risk of nosocomial infection secondary to immunosuppression. Therefore, it should not be administered in all cases, but it should be considered for severe or refractory cases such as with our patient.

Conflict of interest

All authors of this manuscript have no conflicts of interest to declare.

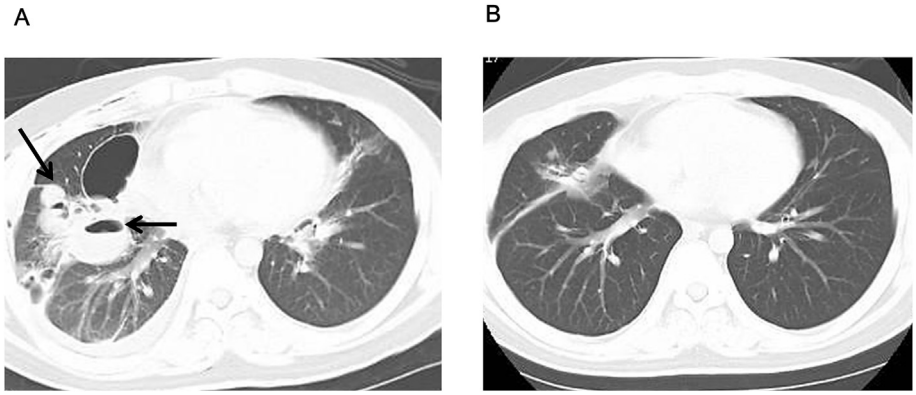

Fig. 4. Two weeks after the initiation of corticosteroid treatment, a computed tomography scan shows pneumatoceles with niveau (arrows) in the right lung accompanied by pleural thickening (A). Two months after the initiation of corticosteroid treatment, a computed tomography scan shows only some bronchiectasis accompanied by pleural and interlobular septal thickening in the right lung (B).

treated with corticosteroid [6]. In our patient, the radiographic abnormality was almost resolved 2 months after the initiation of corticosteroid therapy. The natural history and outcome of lipoid pneumonia are variable. However, the lesions may heal with fibrosis with subsequent loss of lung volume in many cases [8].

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