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Case Report

Endogenous lipoid pneumonia in a cachectic patient after brain injury

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Abstract: Endogenous lipoid pneumonia (EnLP) is an uncommon non-life-threatening inflammatory lung disease that usually occurs in patients with conditions such as lung cancers, primary sclerosing cholangitis, and undifferentiated connective tissue disease. Here we report a case of EnLP in a paralytic and cachectic patient with bronchopneumonia after brain injury. A 40-year-old man experienced a severe brain injury in an automobile accident. He was treated for 1 month and his status plateaued. However, he became paralyzed and developed cachexia and ultimately died 145 days after the accident. Macroscopically, multifocal yellowish firm nodules were visible on scattered gross lesions throughout the lungs. Histologically, many foam cells had accumulated within the alveoli and alveolar walls accompanied by a surrounding interstitial infiltration of lymphocytes. The findings were in accordance with a diagnosis of EnLP. Bronchopneumonia was also noted. To our knowledge, there have been few reports of EnLP associated with bronchopneumonia and cachexia after brain injury. This uncommon pathogenesis should be well recognized by clinicians and forensic pathologists. The case reported here should prompt medical staff to increase the nutritional status and fight pulmonary infections in patients with brain injury to prevent the development of EnLP.

Keywords: Endogenous lipid pneumonia, infection, cachexia, brain injury

Introduction

Lipoid pneumonia (LP) is an uncommon non-infectious inflammatory lung disease that affects humans and animals [1]. LP is characterized by the presence of intra-alveolar lipid and lipid-laden macrophages on microscopy [2]. The usual presentation has an insidious onset as well as nonspecific radiological features and respiratory symptoms such as cough, dyspnea, fever, and chest pain. Thus, it is often difficult to diagnose and is usually found incidentally [3]. The precise incidence of LP is not well known; however, Baron et al. reported a frequency of 1.0-2.5% in a retrospective autopsy study [4]. Based on the lipoid source, LP is categorized as exogenous LP (ExLP) or endogenous LP (EnLP). ExLP classically develops after the inhalation or aspiration of lipids, which mainly occurs in pediatric patients who aspirate mineral oil-based laxatives or older patients with underlying debility, achalasia, reflux, or other neuromuscular disorders of the pharynx and esophagus. However, EnLP is unrelated to such inhalation [5].

EnLP was first described by McDonald in 1949 as "obstructive pneumonitis" in patients with lung neoplasms [6]. Macroscopically, multifocal yellowish firm nodules are seen scattered across gross lesions throughout the lungs; hence, it is also called "golden pneumonia" [7]. EnLP was recently reported in patients with undifferentiated connective tissue disease, primary sclerosing cholangitis, pulmonary alveolar proteinosis, or pulmonary parasitism [8-10]. It is also occasionally found in patients with bacterial or fungal pneumonia [11, 12]. Here we report an uncommon case of EnLP associated with bronchopneumonia and cachexia after brain injury and discuss the potential mechanism in detail.

Case report

Case history

A 40-year-old man experienced an automobile accident on 21 July. He was sent to a local hospital and diagnosed with a severe brain injury. His status plateaued after 1 month of treat-

Figure 1. A. The body presented with skeletization and cachexia; B. Multiple bedsores were seen on the occipital bone, back, hips, heels, and left knee.

ment. However, his right upper and lower extremities were paralyzed. He was neglected by his relatives and did not receive sufficient care other than being fed semifluid rice soup twice daily. Subsequently, weight loss and sacrococcygeal bedsores occurred and the patient's condition deteriorated gradually with severe emaciation, multiple bedsores, and abnormal breath sounds. He ultimately died 145 days after the accident.

Autopsy findings

The body was frozen until a forensic autopsy was performed 3 days after death. Skeletization and cachexia were noted (**Figure 1A**). Multiple severe bedsores were seen on the occipital bone, back, hips, heels, and left knee (**Figure 1B**). Scars were noted on the left temporal and parietal scalp that measured 2.5 cm × 0.3 cm. One old left temporal bone fracture and one fibrous scar measuring 5 cm × 2 cm and 5 cm × 1 cm respectively were seen on the left temporal and parietal lobes. Differing degrees of atheromatous plaque were detected in the left anterior descending, left circumflex, and right coronary arteries. The left and right lungs weighed 300 g and 440 g, respectively. Gross lesions containing multifocal, often subpleural, yellow-white nodules were scattered throughout the lungs (**Figure 2A**, **2B**). The other organs were unremarkable on macroscopic examination.

Histological and toxicological examination

On microscopic examination, glial cells, fibrocytes, foam cells, and necrotic tissue were

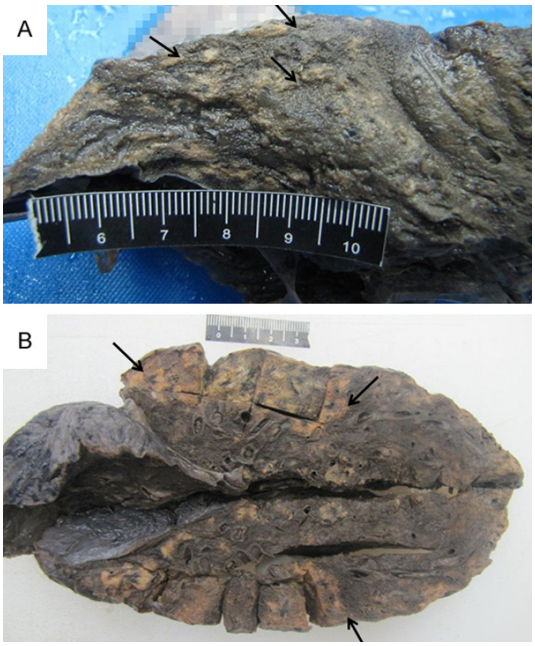

Figure 2. The lungs showed multiple small solid yellow-white foci with an irregular distribution (arrows) over the surface of the lung and in the cut sections.

noted in the corresponding fibrous scar area in the brain. Many foam cells had accumulated in the alveoli and alveolar walls (**Figure 3A**, **3B**), and a mild lymphocytic infiltration was localized around the lesions. Most of the lesions were located under the pleural surface and within the parenchyma, usually within the peribronchial or perivascular regions. Oil red O staining revealed that red-stained fat-storing cells had accumulated within the alveoli and alveolar walls (**Figure 4**). Some small bronchial lumen was blocked by cellular debris and neutrophils (**Figure 3C**), surrounding which the alveoli were diffusely filled with neutrophils (**Figure 3D**). The liver cells showed diffuse atrophy. The toxicology analysis revealed no positive findings.

The cause of death was identified as multiple organ failure after brain injury, and his coronary artery disease might contribute to death.

Discussion

LP is a specific form of pneumonia; however, due to its nonspecific clinical presentation and radiological features, histopathological confirmation is necessary to its diagnosis. In the present case, the pathological finding that lipid-rich foam cells had accumulated within the alveoli and alveolar walls which was consistent with a diagnosis of LP. ExLP was excluded on

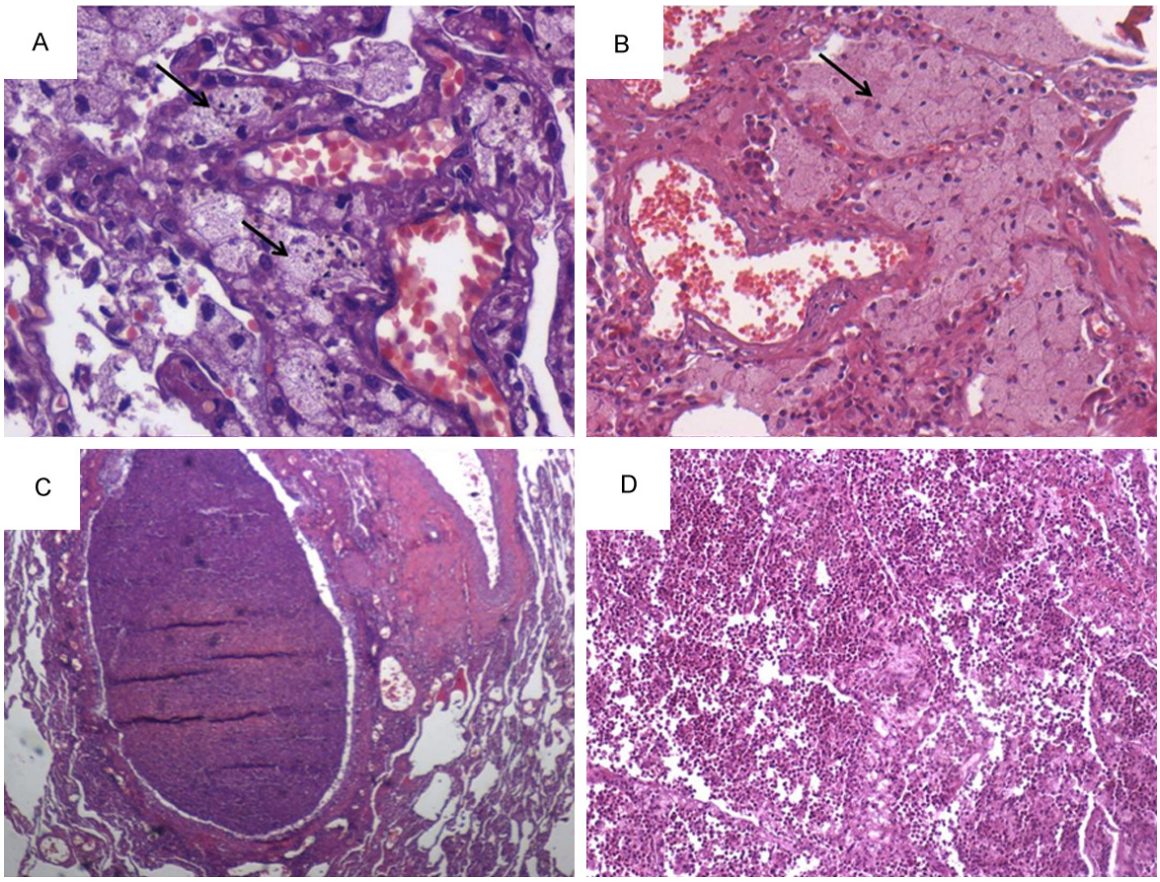

Figure 3. A. Many foam cells had accumulated within the wall of the alveolar (hematoxylin and eosin [H&E] stain, ×400); B. The alveoli was filled with numerous foam cells (H&E stain, × 200); C. Some small bronchial lumen were blocked by cellular debris and neutrophils (H&E stain, × 40); D. Alveoli surrounding the small bronchia were diffusely filled with neutrophils.

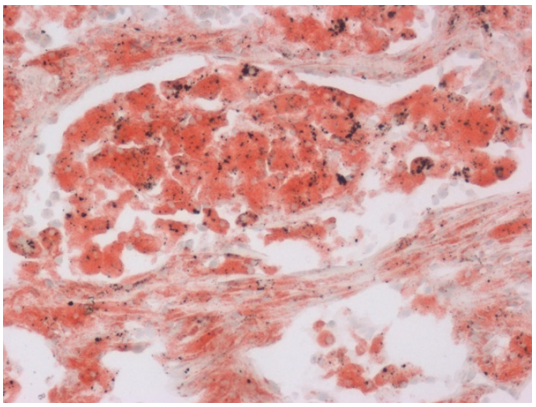

Figure 4. Diffuse red-stained fat-storing cells had accumulated within the alveoli and alveolar walls (Oil red O stain, × 200).

the basis of the fact that the patient had not ingested oily substances. Combined with the autopsy finding that multifocal yellow-white nodules were scattered throughout the lung, a diagnosis of EnLP was made.

EnLP is related to a variety of causes, such as bronchial obstruction, inhalation of irritating dust particles, infection, and lipid metabolism disturbances [13, 14]. In the case described here, the clinical history and the histopathology were indicative of a bronchopneumonia. Thus, EnLP may have been a sequel to infection. Persistent bronchopneumonia can lead to airway blockage and pneumocyte damage consisting of type II pneumocyte hyperplasia and surfactant overproduction [12]. The airway blockage and infection may also result in restrictive pulmonary function and hypoxia. This anoxic tissue injury may stimulate various enzymes such as phospholipases and monooxygenases, which in turn modify low-density lipoprotein within the lung tissue. It has been demonstrated that the modified low-density lipoprotein enhances lipid uptake by alveolar macrophages [14]. Thus, infection leading to tissue damage and anoxia must be considered as a risk factor for EnLP. At the same time, EnLP also further aggravates pulmonary function.

Our patient also had cachexia, which may have contributed to the formation of EnLP. Currently, the correlation between EnLP and malnutrition remains unknown. Costa et al. reported a similar case of EnLP in a malnourished parrot [7]. In that case report, the parrot experienced an inbalanced diet. Moreover, Beaver et al. demonstrated that a protein-deficient cirrhogenic diet is linked to EnLP in laboratory rats [15]. Thus, the inappropriate diet in the pathological process of malnutrition maybe a high risk factor resulting in EnLP. Interestingly, the patient in our case also experienced an imbalanced diet characterized by low protein and energy contents over the longer term. The skeletization of the victim demonstrated that he had a remarkably insufficient energy supply that may have contributed to the development of EnLP.

Conclusions

The EnLP in our patient was considered an incidental postmortem finding. Although EnLP is not life-threatening, it could aggravate an individual's pulmonary function. To our knowledge, EnLP associated with bronchopneumonia and cachexia after brain injury is very rare. This rare pathogenesis should be well recognized by clinicians and forensic pathologists. The case reported here should prompt medical staff and the victim's relatives to attend to patients after severe brain injury. It is important to increase the nutritional status and fight pulmonary infections in patients with brain injury to prevent the development of EnLP.

Disclosure of conflict of interest

None.

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