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Case Report

Pulmonary lymphoepithelial cyst with no prior HIV infection: A case report☆,☆☆

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ABSTRACT

Most lymphoepithelial cysts (LECs) occur in the salivary glands and are considered one of the autoimmune syndromes caused by the human immunodeficiency virus (HIV). In this report, we present a case of pulmonary LEC without prior HIV infection, paying special attention to radiographic features. A chest radiograph revealed an oval mass with a smooth surface, localized in the left lower lung field, which was in direct contact with the diaphragm. Computed tomography showed an oval homogenous mass with a smooth surface in the lower left lobe. Further, magnetic resonance imaging demonstrated that the mass was a homogeneous internal structure with a smooth surface and a slightly high signal in T2-weighted images and a slightly low signal in T1-weighted images. Surgical resection was performed, and pathological examination confirmed the diagnosis of a pulmonary LEC. To the best of our knowledge, no cases of pulmonary LECs without prior HIV infection have been reported in the literature to date, therefore, the case presented here is considered very rare and informative.

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Introduction

Most lymphoepithelial cysts (LECs) occur in the salivary glands and are considered one of the autoimmune syndromes caused by the human immunodeficiency virus (HIV) [1]. Although many etiological theories are underlying LECs, no definitive consensus regarding their causes has yet been established. Structurally, a LEC is lined with squamous or columnar epithelium, and the wall is formed by lymphoid tissue

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with a germinal center. Pathologically, it resembles a branchial cyst. Lung LECs is extremely rare, and there are only a few published case reports [2–4]. However, there are no consistent reports on their radiographic features. Parotid LECs and pancreatic LECs are usually displayed as masses with a contrast effect on the wall, with varying internal absorption values and signal intensity dependent on the internal composition. Here, we describe a LEC in the lungs of a patient who has not previously had an HIV infection. We have not been able to find a single case of pulmonary LECs without prior HIV infection, and the case presented in this report is considered very rare.

Case presentation

A 46-year-old non-smoking HIV-negative woman with a medical history of *Helicobacter pylori* infection presented with mild chest pain and recurring cough. A chest radiograph revealed a solid tumor of 3 cm in size with a smooth surface in the left lower lung field with no calcification component. The tumor was in direct contact with the diaphragm (Fig. 1). Computed tomography (CT) of the chest revealed an oval uniform mass with a smooth surface of approximately 3 cm in size in the left lower lobe (Fig. 2A). Contrast-enhanced CT (CE-CT) showed a homogenous non-enhancing cystic mass (Fig. 2B). There was no internal calcification or fatty component. Magnetic resonance imaging (MRI) also revealed a 3 cm diameter formation with a smooth surface. In T2-weighted images, the mass had a homogeneous high signal intensity slightly lower than that of free water, and in T1-weighted images, it had a homogeneous low signal intensity slightly higher than that of free water, which indicated the presence of internal protein inclusions (Fig. 3). Both the diffusion-weighted image and the apparent diffusion coefficient map showed a high signal, with no lesions of low signal intensity. Based on the radiographic features alone, bronchogenic cyst or hamartoma are also suggest diagnosed differentially. Based on the location of the cyst, a solitary fibrous tumor of the pleura was also identified.

The patient underwent surgery with partial resection using the method of video-assisted thoracoscopic surgery. Intraoperatively, it was revealed that the mass was not in contact with the pleura. The tumor was yellow on the surface, and a gross pathological examination revealed a unilocular cystic mass filled with mushy material (Fig. 4B). No significant components were found in the lumen (Fig. 4A). Microscopically, the cyst was lined with stratified squamous epithelium, and lymphocytic infiltration with lymphoid follicles was apparent in the cyst wall (Fig. 5). The presence of normal lung tissue outside the tumor indicated that the lesion was of pulmonary rather than pleural origin. The tumor was filled with necrotic material comprised of keratocytes and neutrophils. Taken together, these pathological findings led to the diagnosis of a LEC of the lung.

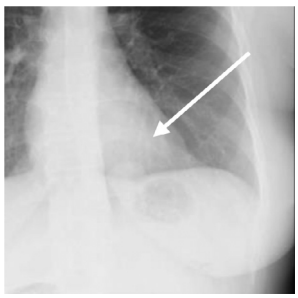


Fig. 1 – Chest radiograph showed a tumor in the left lower lung field.

Discussion

LECs are considered part of the autoimmune-like syndrome caused by HIV and are well described in connection with the pathology of the salivary glands. Pulmonary LECs are a rare cystic disease, and only 3 cases have been reported in the literature [2–4]. All 3 patients presented in these reports had a history of HIV infection. To the best of our knowledge, the case presented here is the first case of non-HIV-related pulmonary LECs. Although it is known that LECs also tend to occur in the pancreas, we were not able to find any publications on pancreatic LECs associated with HIV.

There are 2 main theories behind the development of the parotid gland LECs. One of them is the branchial apparatus cleft theory, which states that cysts develop from the remnants of the branchial cleft since they arise in the area of the embryonic gill apparatus. The other is the parotid gland inclusion theory, which suggests that cysts arise as a result of cystic changes in the parotid gland, subsequently getting trapped in the upper cervical lymph nodes during embryonic development [5]. If the first theory is confirmed, the formation is called a LEC; if the second, then it is called a branchial cyst. In addition, there are several theories behind the development of pancreatic LECs. Those can arise from (a) squamous metaplasia of the pancreatic ducts with subsequent cystic transformation, (b) ectopic remnants of the branchial cleft cyst that became displaced and fused with the pancreas during embryogenesis, (c) epithelial remnants within peripancreatic lymph nodes, and (d) ectopic pancreatic tissue included in the peripancreatic lymph node [6].

The number of documented cases of pulmonary LEC is small, and further accumulation of cases is needed. Pathologically, the cyst was lined with squamous or columnar epithelium, and the wall was formed by lymphoid tissue with a germinal center. The pathological findings were consistent with those of previous reports. The absence of skin appendages on the wall distinguished that formation from an epidermoid cyst. On the other hand, the internal contents of the cyst vary from case to case, and all reports of pulmonary LEC cases documented that the cyst was filled with clear serous fluid. In the case described here, necrotic material with keratinization and neutrophil infiltration was observed. The internal contents of both the pancreatic and the parotid LECs were reported to be cheese-like or serous, and this case appeared to seemed to match the features corresponding to LECs. The mass did not show a clear contrast effect on the CE-CT, which suggests that it was not a solid tumor. Non-CE-CT showed a slightly high mass density, and T1-weighted images showed a slightly high

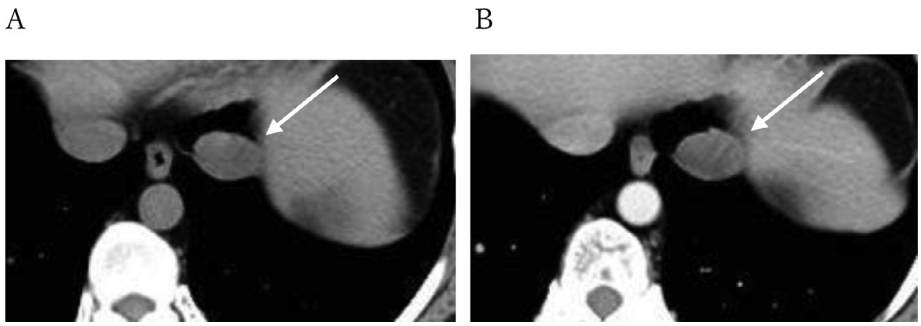


Fig. 2 – (A) Non-contrast-enhanced computer tomography (Non-CE-CT) revealed an oval homogenous mass with a smooth surface of slightly low density. The mass was approximately 3 cm and was localized in the left lower lobe (arrow). No internal calcification or fatty component was detected. (B) CE-CT revealed a non-enhancing cystic mass.

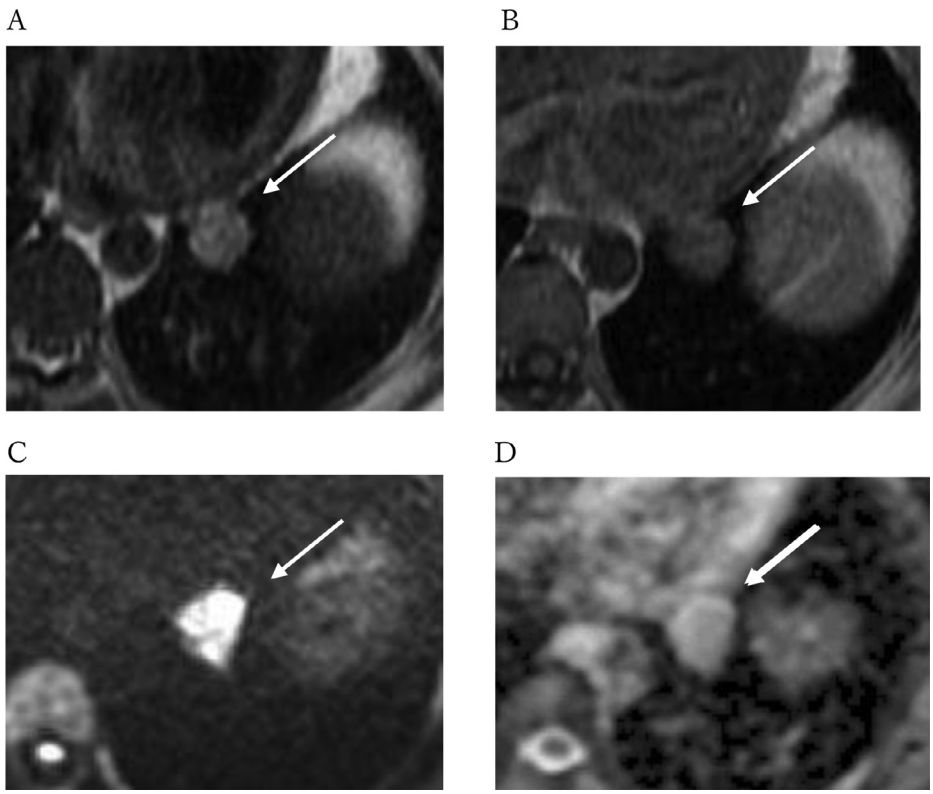


Fig. 3 – (A) T2-weighted images showed a homogeneous mass with a slightly lower signal intensity than that of free water. (B) T1-weighted images showed a homogeneous mass with a slightly higher signal intensity than that of free water, indicating the inclusion of protein components. (C and D) Diffusion-weighted images (DWI) showed a high-intensity signal, with no reduction on the apparent diffusion coefficient (ADC) map.

signal, both indicative of a cystic tumor rich in protein components. The pathological examination revealed a cystic tumor containing necrotic material, which was believed to correspond to the findings of CT and MRI.

In a report of a LEC in the parotid gland of an HIV-negative patient, the contents were described as cheese-like, which was similar to our case [7]. Pathological characteristics of LECs in HIV-positive and HIV-negative patients tend to be similar, but the pathogenesis may be different. Although it was previously described that one of the typical radiographic features of a LEC is a contrast-enhanced cyst wall [8], in this case, CE-CT showed a cystic lesion with no contrast enhancement. One previous case of an HIV-negative patient also reported the presence of the parotid gland LEC that had no cyst contrast enhancement on CE-CT [7].

In previous cases, it was reported that the pulmonary LEC was localized near the hilum of the lung [2–4]. In the case presented here, the LEC was in contact with the diaphragm, which appears to be different from the location indicated in previous reports. There is no consensus on the standard of care for LEC in non-HIV patients. Treatment of parotid lymphoepithelial lesions in HIV-positive patients involves clinical

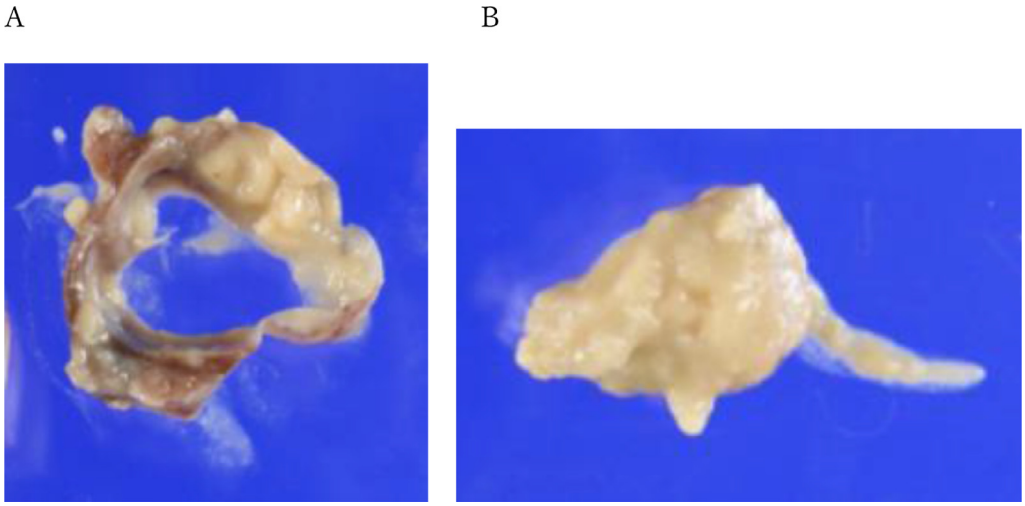


Fig. 4 – A surgical specimen, (A) in the lumen of which no significant components were found, and (B) the lumen of which was filled with mushy material.

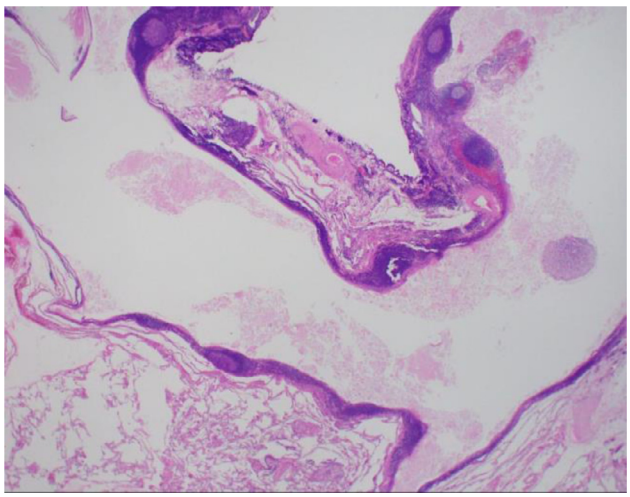


Fig. 5 – Microscopic section of a specimen showing a cyst covered with stratified squamous epithelium. Lymphocytic infiltration with lymphoid follicles was prominent in the cyst wall (HE x100).

monitoring and antiviral therapy [9]. If only the aspiration of the internal contents fluid is considered, the disease tends to recur following the procedure. Sodium morrhuate injections, surgery, and radiation therapy may be considered, but they should be limited due to concomitant side effects [9,10].

Conclusion

We report a rare case of pulmonary LEC, with an emphasis on CT and MRI findings. The radiographic features of the lesion were consistent with gross pathological findings. To the best of our knowledge, this is the first case of a pulmonary LEC not associated with HIV.

Patient consent

Informed consent was obtained from the patient.

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