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HIV-associated (non-thymic) intrathoracic lymphoepithelial cyst in a child

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Abstract An unusual case of a juxtabronchial lymphoepithelial cyst in an HIV-positive child with post-obstructive pneumonia is presented. The pathogenesis and similarity with parotid lymphoepithelial cysts is discussed.

Introduction

Parotid lymphoepithelial cysts (LEC) have been well described in adults and children with human immunodeficiency virus (HIV), type 1 [1]. We present an unusual case of an HIV-positive child with recurrent post-obstructive pneumonia secondary to a juxtabronchial LEC. To our knowledge, this is the first report of a intrathoracic non-thymic LEC in a child with HIV infection.

Case report

A 10-year-old HIV-positive boy presented to the emergency room with a right upper lobe infiltrate and a 3-cm, right suprahilar mass interpreted as adenopathy. Of note, the patient had no parotid enlargement and no history of lymphocytic interstitial pneumonia. The patient was treated for bacterial pneumonia and tuberculosis despite a negative tuberculin test. The pneumonia cleared and the patient was lost to follow-up.

Three years later the patient was readmitted with an infiltrate in the right upper lobe (RUL) (Fig. 1). CT scan demonstrated a 3 × 2-cm cystic mass in the right suprahilar region adjacent to the apical branch of the RUL bronchus (Fig. 2), and a RUL infiltrate with bronchiectasis. The patient was treated with antibiotics and was again lost to follow-up. Six months later he returned with a recurrent RUL pneumonia and a right suprahilar mass. After resolu-

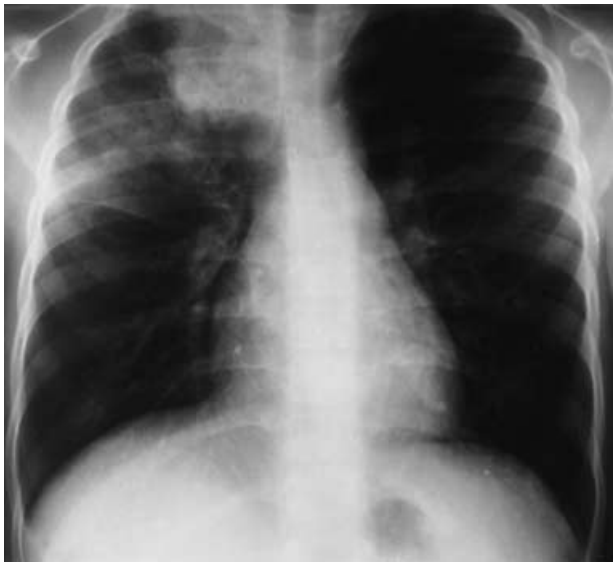

Fig. 1 Frontal chest radiograph demonstrates a RUL pneumonia. The right suprahilar mass is obscured by the lung disease

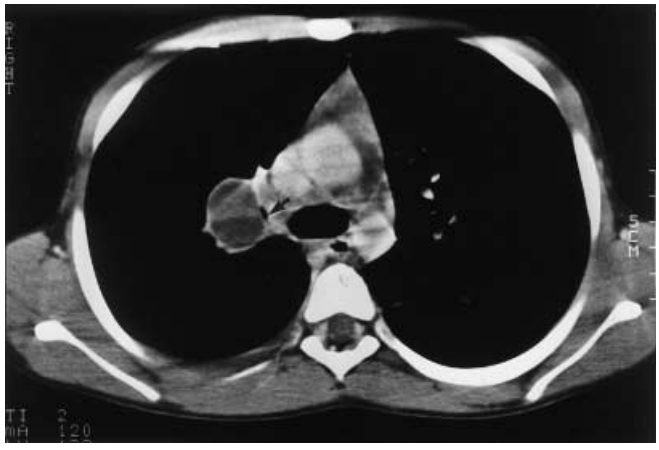

Fig. 2 CT scan of the chest demonstrates a low-density mass in the right suprahilar region inseparable from the mediastinum. Note the close proximity of the mass to a portion of the right upper lobe bronchus (*arrow*)

tion of the pneumonia following antibiotic therapy, the patient underwent a thoracotomy. At surgery, 10 cm^3 of turbid fluid was aspirated from the extrapulmonary, juxtabronchial mass. Because the mass was grossly adherent to the RUL pulmonary artery and pulmonary vein and compressed the RUL bronchus, it was partially excised and marsupialized. Pathology revealed lymphoid and fibrous tissue with respiratory and squamous epithelium in the cyst wall consistent with a LEC. No inclusion bodies were identified.

Discussion

Lymphoepithelial cysts of the salivary glands, first described in 1985 [2], have been well described in adults and children seropositive for human imunodeficiency virus type 1 [1].LECs occur most commonly in the parotid gland, although they have also been described in the submandibular gland, tonsils, thyroid gland and pancreas, and vary from simple cysts to complex cystic and solid masses. They are considered benign reactive lesions [3]. LECs are often associated with diffuse lymphocytosis syndrome, (DILS) which is characterized by CD8 lymphocytosis, visceral CD8 lymphocytic infiltrate (most commonly of the lung), bilateral parotid swelling and cervical adenopathy [4]. Of note, our patient demonstrated none of these signs. Pathologically, the cysts are characterized by lymphoid and epithelial cells as well, as germinal centers in the cyst wall [4]. They may serve as a reservoir of HIV particles [5].

Intrathoracic LEC is an unusual entity and we could find only a single report of an intrathoracic LEC occurring in a 35-year-old woman with HIV who presented with bilateral lung masses [6]. As in our case, the masses were perihilar in location, and at operation the left lung mass was in close proximity to the lingular bronchus. It has been postulated that, like parotid LEC that develop in noninflammatory lymphoid tissue within the parotid gland, or in intra- or paraparotid lymph nodes [7], intrathoracic LECs develop in peribronchial lymph nodes or in noninflammatory peribronchial lymphatic tissue [6]. The development of the LEC in close proximity to the apical segment of the RUL bronchus in our patient contributed to recurrent post-obstructive pneumonia.

Mandel and Hong [4] has proposed that parotid LEC may develop within extraglandular infiltrative lymphoid tissue. Our patient had no history of pulmonary lymphoid hyperplasia. However, given the association between parotid LEC and lymphoid infiltration of the lung, the occurrence of intrathoracic LEC is not entirely surprising.

The radiographic and histologic findings of intrathoracic LEC and parotid LEC are similar to those of HIV-related multilocular thymic cysts (MLTC). MLTC have been described in HIV in both children and adults, often in the setting of DILS [8, 9]. It is likely that intrathoracic LEC, parotid LEC, and MLTC all represent similar manifestations of DILS. Pathologically, they are believed to result from cystic transformation of duct epithelial structures induced by the reactive lymphoid hyperplasia accompanying HIV infection [8–10].

In summary, although there are many more common causes of lung masses in a HIV-positive child, including infection and neoplasm, the diagnosis of intrathoracic LEC should be entertained if the child presents with one or more central, cystic pulmonary masses adjacent to the tracheobronchial tree. If one extrapolates from the clinical experience with parotid LEC and MLTC (both of which have a benign course), it may be reason-

able to observe rather than surgically remove intrathoracic LEC if the child is asymptomatic and no complicating features such as post-obstructuve pneumonia develop. Close radiographic follow-up can be performed. A solid lesion should never be assumed to be an intrathoracic LEC.

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