

Case report - Thoracic oncologic Extrapleural pneumonectomy for pyothorax-associated lymphoma

Semih Halezeroglu^{a,*}, Atif Akcevin^b

^aDepartment of Thoracic Surgery, Acibadem Maslak Hospital, Istanbul, Turkey

^bDepartment of Cardiovascular Surgery, Kadikoy F. Nightingale Hospital, Istanbul, Turkey

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Abstract

Pyothorax-associated lymphoma (PAL) is a non-Hodgkin's lymphoma that occurs in the pleural cavity of patients with a long-standing history of pyothorax. Here, we report a left pleuropneumonectomy performed for PAL in a 76-year-old male patient with a 60-year history of pyothorax due to tuberculosis.

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Keywords: Pyothorax-associated lymphoma (PAL); Pleuropneumonectomy; Chronic pyothorax; Tuberculosis; Lung resection

1. Introduction

Pyothorax-associated lymphoma (PAL) is a non-Hodgkin's lymphoma of exclusively B-cell phenotype in the pleural cavity of patients with a long-standing history of pyothorax and is strongly associated with Epstein-Barr virus (EBV) infection. It has been rarely reported from a country other than Japan [1, 2]. Although surgery has a limited role in other types of lymphomas, surgical resection is a curative treatment for PAL [3, 4]. However, use of a pleuropneumonectomy for treatment of this disease has not been described in the English literature.

2. Case report

A 76-year-old man was admitted with persistent cough, blood-stained sputum, fever, fatigue, and weight loss. He reported that his present illness had started approximately a year ago, manifesting with acute bronchitis symptoms – a dry cough was followed by a productive cough. His medical history included an artificial pneumothorax for tuberculosis and a prostatectomy for prostate cancer, performed 60 and 10 years ago, respectively.

The auscultation revealed no breath sounds from his left chest. Laboratory test results were as follows: C-reactive protein (CRP), 210 mg/l; erythrocyte sedimentation rate (ESR), 127 mm/h; leukocyte count, 16.000/mm³; and neuron-specific enolase (NSE), 41 ng/ml. Laboratory studies and cultures for bacterial, viral, and fungal infections were all negative. A chest X-ray revealed total opacity of the left side, and computed tomography (CT) of the chest showed a large, calcified pleural pouch, a 7×5 cm central

mass lesion and a destroyed lung in the left chest (Fig. 1a). No endobronchial lesion was observed during fiberoptic bronchoscopy. A pathological diagnosis could not be made with either a transbronchial or CT-guided trans-thoracic fine needle aspiration of the central tumour. Because the disease was not identified pathologically, only supportive treatments were initiated. The patient continued to lose weight, and his general status deteriorated. A CT-scan of the thorax performed 5 months after the initial hospital admission revealed that the tumour in the left chest had enlarged significantly and, invaded the pyothorax pouch (Fig. 1b). The only finding on positron emission tomography was increased fluorodeoxyglucose uptake by the central tumour (Fig. 2). Magnetic resonance imaging showed no brain lesions. Forced expiratory volume in the first second in pulmonary function test and the carbon monoxide diffusing capacity were 51% and 85% of predicted, respectively. His maximal oxygen consumption rate was 15 ml/kg/min. The cardiac ultrasound was normal.

Physical and radiographic examinations of the patient suggested a progressive, yet, localized malignant tumour developing in chronic inflammation in the left hemithorax. We discussed with the patient and his family possible outcomes of two treatment options, observation with supportive treatment or pleuropneumonectomy, and they opted for the surgery.

A left posterolateral thoracotomy was performed by excising the fifth and sixth ribs. En-block extrapleural resection of the empyema pouch and destroyed lung that contained the tumour was performed with lymph node dissection. The pathology of the specimen was EBV positive diffuse large B-cell lymphoma, and the lymph nodes were not involved.

The postoperative course was uneventful, and the patient was discharged from hospital on the 8th postoperative day. At the last follow-up, 12 months after surgery, his perform-

*Corresponding author. Department of Thoracic Surgery, Acibadem Maslak Hastanesi, 34398 Istanbul, Turkey. Tel.: +90 5322660234.

E-mail address: semihh@atlas.net.tr (S. Halezeroglu).

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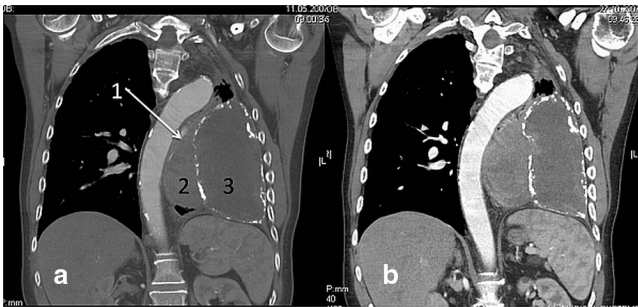


Fig. 1. Computed tomography images of the chest. (a) On admission, 1, destroyed left lung; 2, central tumour; and 3, calcified pleural pouch were seen; (b) Five months after the initial admission, the tumour had enlarged and invaded the pleural pouch.

ance had significantly improved, his body weight had normalized, and CRP, ESR, leukocyte, and NSE levels were normal.

3. Comment

PAL was described as a distinct clinicopathologic entity in 1987 [1]. The disease is more common in males and in patients with a 20- to 64-year history of pyothorax [1–6]. The main causative factor for PAL has been identified as artificial pneumothorax for treatment of tuberculosis and subsequent long-standing pyothorax [1–6]. In about 70% of PAL cases, patients were reported as positive for EBV infection [6]. NSE, which is increased in the majority of patients, is an important parameter in the follow-up of response to the treatment [3, 4].

PAL has been reported primarily in Japan where it is diagnosed in ~2% of patients with chronic pyothorax [1]. Some authors correlated the higher prevalence in Japan with the frequent use of artificial pneumothorax or with genetic factors [6]. Only a few case reports are available in literature from outside of Japan, and, all these reports describe the diagnosis of this rare disease with biopsy or debridement [7]. There has been no curative intend pulmonary resection reported in the English literature.

Optimal management of PAL has not been determined. Almost 80% of patients with PAL receive chemotherapy and/or radiotherapy, but the overall prognosis is poor [3, 6]. The estimated 1-, 3-, and 5-year survival rates were reported as 48.6, 27.0, and 21.6%, respectively [3, 6]. Two large case series showed that pleuropneumonectomy, either alone or in combination with chemotherapy and/or radiotherapy, was curative [3, 4]. However, as most patients with PAL are elderly and/or have comorbidities, surgery



Fig. 2. Positron emission tomography showed increased fluorodeoxyglucose uptake by the central tumour.

can be performed only in a certain group of patients who have good performance status.

In such cases, the destroyed lung, pyothorax pouch and the tumour are resected en-block by dissecting them vigilantly from the chest wall, mediastinum, and diaphragm in an extrapleural fashion. Pleuropneumonectomy and its postoperative care are challenging in a patient with chronic pyothorax. For this reason, a surgeon who attempts a surgical resection in a patient with PAL needs to be experienced in treating patients with chronic inflammatory pleuropulmonary diseases.

In conclusion, considering PAL in differential diagnosis of patients with a long-standing history of pyothorax will help distinguish it from other malignancies. Pleuropneumonectomy might be curative in selected patients with good performance status who have the PAL confined to the primary tumour site.

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