Postpneumonectomy-like syndrome after chemoradiation therapy for lymphoma

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Postpneumonectomy syndrome (PPS) is a rare complication of pneumonectomy due to an excessive mediastinal shift producing compression of the main bronchus or a lobe bronchus on the aorta or the spine. We report an exceptional case in which an extreme mediastinal shift was due to fibrosis and complete atelectasis of the left lung, as a complication of chemoradiation treatment for recurrent mediastinal Hodgkin’s lymphoma. This condition, associated with a further recurrence of the disease, indicated a postpneumonectomy-like syndrome.

Key words: complication, Hodgkin’s lymphoma, pneumonectomy, therapy

Introduction

Postpneumonectomy syndrome (PPS) is a rare condition characterised by dyspnea and stridor due to an extreme mediastinal shift and bronchial compression of the residual lung after pneumonectomy. Even though a certain degree of lung toxicity and fibrosis are frequently described after chemotherapy and/or chest irradiation, no cases describing such a clinical picture in the absence of pneumonectomy have been reported in the literature. We report a unique case of complete left lung collapse and severe mediastinal deviation due to chemoradiation therapy for recurrent Hodgkin’s lymphoma (HL). This condition, in association with a further recurrence of the disease at a mediastinal level, caused a postpneumonectomy-like syndrome.

Case report

A 44-year-old woman presented with severe progressive dyspnea. Patient history commenced in 1987 with a diagnosis of HL of the supraclavicular nodes (stage IIB), for which she received eight cycles of chemotherapy with MOPP (meclotriline, oocarbide, procarbazine, prednisone) and ABVD (doxorubicin, bleomycine, vinblastine, docarbazine). In May 1988, high-dose chemotherapy (not concluded) and left mediastinal irradiation were performed for a mediastinal recurrence of her disease. In 1989, a third line of chemotherapy with CEP (lomustine, etoposide, prednimustine) for six cycles was administered for a second relapse at the mediastinal lymph nodes, and a complete response was achieved. One year later a chest X-ray demonstrated atelectasis of the left lower lobe and initial mediastinal shift that progressed over the following years (Figure 1). The patient remained in a good state of health for the next 10 years.

In March 2000, the patient complained of a progressive dyspnea lasting several months, with recurrent crisis of severe respiratory distress. A computed tomography (CT) scan of the chest showed an extreme mediastinal shift, a complete left lung atelectasis and a hyperdistension of the right lung (Figure 2). An image of compression of the right main bronchus was clearly evident on a three-dimensional reconstruction of the airways (Figure 3). A solid lesion of $8 \times 5$ cm was found in the left upper hemithorax, in a completely collapsed, non-
functional lung. A biopsy diagnosed a recurrence of HL and a chest magnetic resonance imaging scan confirmed the CT findings. A pulmonary function study demonstrated a moderate obstructive-restrictive syndrome (forced expiratory volume in 1 s, 50.6%; functional volume capacity, 67%). A conservative treatment with corticosteroids was able to immediately improve the respiratory difficulty. Medical treatment with CHEVPP (chlorambucil, doxorubicin, vincristine, procarbazine, prednisone) was undertaken to decrease the left lung lesion and to improve the dyspnea further. One year on from the diagnosis of postpneumonectomy-like syndrome, the patient is alive with a stable respiratory function, and has suffered no more episodes of acute distress since the medical treatment. At present, treatment is ongoing for a vertebral deposit of the disease.

Discussion

Lung toxicity and fibrosis are well described complications of systemic chemotherapy or chest irradiation. In the present case, however, extreme fibrosis, collapse of the left lung and a severe mediastinal shift occurred as a consequence of radiochemotherapy for recurrent HL. The novel recurrence of the disease caused a particular condition characterised by compression of the main bronchus and a clinical picture comparable to a well known complication of pneumonectomy, PPS.

Figure 2. Computed tomography scan of the chest: clockwise rotation of the mediastinum and great vessels. The trachea is to the left of the spine, and the right main bronchus is cross-anterior to the thoracic spine. A mass of ~6 cm is visible in the upper part of the left lung.

Figure 3. Three-dimensional reconstruction of the airway demonstrates compression of the right main bronchus. The left lung is not visible due to its complete atelectasis.
PPS is a very rare complication, particularly observed in children or young adults, occurring between a few months and many years after pulmonary resection [1], characterised by progressive dyspnea and stridor not justified by the pneumonectomy and due to bronchial compression. Diagnosis is usually achieved using a CT scan and bronchoscopy. Successful management varies according to different experiences [2, 3].

In the present case, a postpneumonectomy-like syndrome developed in the absence of pneumonectomy. The mediastinal deviation was the consequence of left hemithorax fibrosis due to mediastinal irradiation, and lung toxicity due to chemotherapy. The clinical picture of progressive dyspnea, stridor and airway compression are explained by the combination of two events: the extreme mediastinal shift and the recurrence of HL. Although a prompt surgical treatment is usually necessary with PPS to avoid further complication of the syndrome or to reduce the chance of potential lethal bronchial malacia, in this case a non-surgical approach was adopted to cope with the neoplasm that concurred with the airway obstruction. This treatment led to sudden improvement of the clinical picture.

In conclusion, we present an exceptional case of extreme mediastinal shift associated with complete left lung collapse and controlateral bronchial stenosis as a consequence of a chemoradiation treatment. Salvage chemotherapy proved effective in reducing bronchial stenosis. The term post-pneumonectomy-like syndrome is introduced to underline the similarity of the present case pathogenesis and clinical presentation with those of a well known and rare complication of pneumonectomy.

References