Chronic Inflammation-Related Diffuse Large B-Cell Lymphoma Around the Area of Thoracotomy After Decortication

Dekortikasyon Sonrası Torakotomi Yerinde Kronik Enflamasyon ilişkili Diffuz Büyük B Hücreli Lenfoma

Torakotomi Yerinde Lenfoma / Lenfoma Around the Thoracotomy Area

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Özet

Torasik bölge tümörlerinin %5'ini göğüs duvarı selim ve habis tümörleri oluşturur. Primer göğüs duvarı tümörlerinin ise %5'den az bir kısmını lenfomalar oluşturur. İki yıl önce sol plevral dekortikasyon ameliyatı geçiren 63 yaşında erkek hasta kliniğimize sol göğüs ağrısı şikayeti ile başvurdu. Radyolojik olarak operasyon sahasında kitle lezyonu tesbit edilen hastaya yapılan iğne biyopsisi sonucunda Ewing/PNET veya akciğer kaynaklı tümör olarak düşünülmesi üzerine hasta opere edildi. Operasyon sonrası patolojik incelemede kronik enflamasyon ilişkili diffüz büyük B hücreli lenfoma saptandı. Cerrahi skar zemininde iki yıl gibi erken bir dönemde gelişen kronik enflamasyon ilişkili diffüz büyük B hücreli lenfoma olgusuna literatürde nadir rastlandığı için radyolojik, operasyonel ve patolojik görüntüleri ile sunmayı amaçladık.

Anahtar Kelimeler

Göğüs Duvarı; Kronik İnflamasyon; Diffüz Büyük B-Hücreli Lenfoma

Abstract

Chest wall tumors consist 5% of all tumors in the thorax. Lymphomas compose of less than 5% of all primary chest wall malignancy.Sixty three years old patient who had an operation for pleural thickness two years ago admitted with complaint of left-sided chest pain.Following the detection of mass lesion radiologically at the place of previous operation area, the patient was operated based on needle biopsy result suggesting Ewing /PNET or pulmonary originated tumor. After the operation, pathological examination confirmed chronic inflammation-related diffuse large B-cell lymphoma. Since it has been rarely reported in the literature, we aimed to present the case of chronic inflammation-related diffuse large B-cell lymphoma developed within such a short time as two years on the ground of surgical incision scar tissue together with our radiologic, surgical, and pathological findings.

Keywords

Chest Wall; Chronic Inflammation; Diffuse Large B-Cell Lymphoma

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Introduction

Benign and malign tumors of the chest wall consist of 5% of all tumors in the thorax. The chest wall tumors can be classified into four groups as follows: primary tumors, local tumor invasion of neighboring organs, metastatic lesions, and non-neoplastic masses. Lymphomas compose of less than 5% of all primary chest wall malignancy [1]. There are limited number of case reports diagnosed with primary malign lymphoma originated from pleura, rib, and sternum [1-2]. Herein, we reported a very rare case of chronic inflammation-related diffuse large B-cell lymphoma developed within two years on the ground of surgical incision scar tissue together with our radiologic, surgical, and pathological findings.

Case Report

Sixty-three year old male patient applied with left-sided chest pain. He undergone decortication for left-sided pleural thickness two years ago. And, the operation material had been diagnosed as empyema and chronic pleuritic inflammation without any sign of malignancy histopathologically. Physical examination was unremarkable except painful swelling at the line of surgical incision scar on the left lateral inferior chest wall that developed two years after the previous operation. Superficial ultrasonography examination of the related region revealed a solid soft tissue tumor in left lateral chest wall sized 88x53 mm, consisting of cystic areas suggesting chronic hematoma at first glance. In computed tomographic examiation, a lobulated solid mass sized 10x7.5 cm, infiltrating the neighboring thoracic muscles and destructing the neighboring ribs with extension towards intrathoracic space and subcutanous region was detected at left inferior hemithorax. Positron emission tomography (PET-CT) showed mean uptake of 5.5 SUV(max) at the lesion (Figure 1).

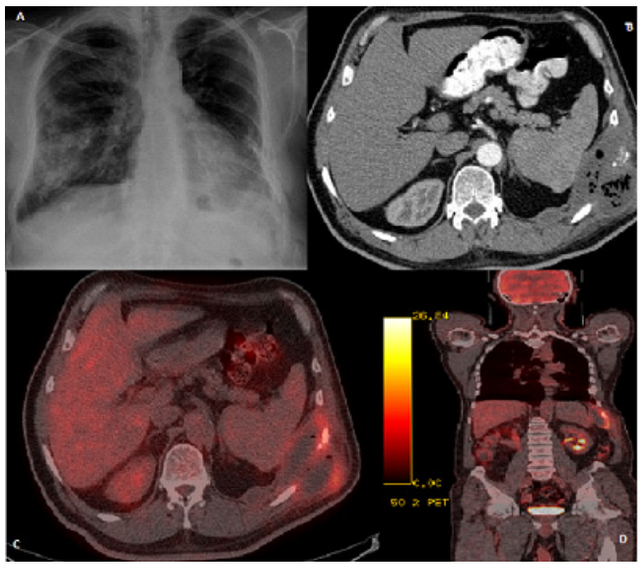

Figure 1. Fracture line at 8th rib on chest X-ray(A), The appearence of the mass including necrotic regions with bone desrtruction on thorax CT (B), The mass with mean uptake of 5.5 SUVmax at left thoracic wall on PET-CT (C,D).

Then a tru-cut needle biopsy was performed. Under light microscope, the biopsy showed small amount of viable tumor cells with small round nuclei (left) and scant cytoplasms suggesting of small round blue cell tumors (Figure 2) (small cell carcinoma,

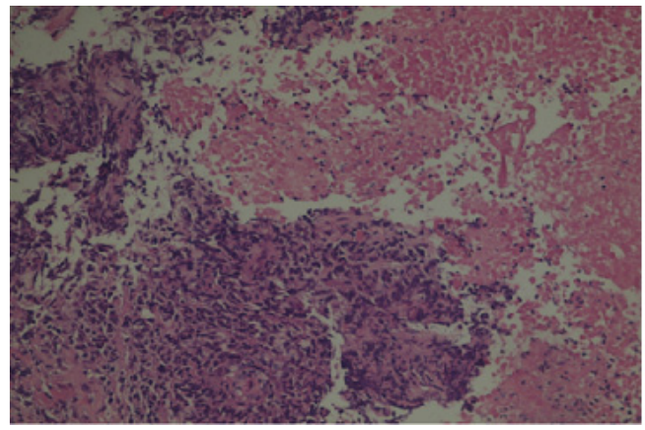

Figure 2. Groups of viable tumor cells with small round nuclei (left) and scant cytoplasms in the necrotic areas (right), (Hematoxylin and eosin stain, x100).

lymphoma, melanoma, Ewing/PNET, and rhabdomyosarcoma, etc.) in excessive necrotic tissues. Immunohistochemical staining was applied in order to obtain the exact diagnosis. The tumor was found to be positive for TTF-1 and CD99. Chromogranin A, synaptopysin, pancytokeratin, SMA, desmin, CD34, HMB-45, CD3 and CD79a were negative. CD20 was ineffective. Then the histopathological and immunohistochemical findings were reported as suggesting of Ewing/PNET or a tumor originated probably from the lung.

Then, the operation decision for total removal of the mass was given upon preoperative routine cardiologic and anesthesia evaluation. At the operation, en bloc resection from 7th rib to 12th rib was performed. Since the mass was observed to be extending toward spleen, partial wedge resection was performed with the help of cautery by the general surgeon. The part of diaphragma involving the mass was resected. Lung tissue neighboring the mass was removed with wedge resection. The tissue defect in chest wall and diaphragma formed after the resections were reconstructed with composite mesh (Figure 3).

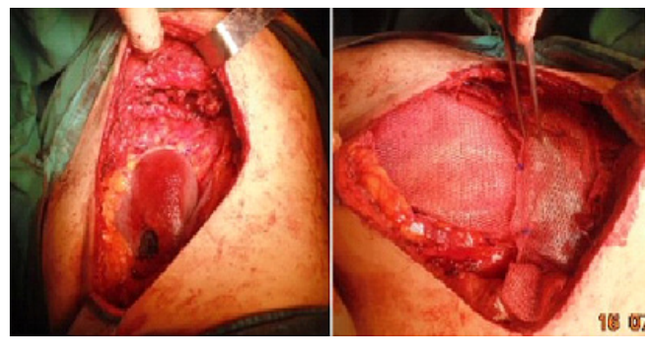

Figure 3. Tissue defect of the patient due to extensive resection of thoracic and abdominal wall was repaired with composite mesh.

Under the light microscope, the tumor of the chest wall was composed of extensive necrosis and small amounts of viable small round blue tumor cells as in the tru-cut biopsy. Immunohistochemically the tumor cells showed positivity for CD20, CD21, an indicator of B cell. In addition, it showed positivity for both Epstein-Barr virus (EBV) immunohistochemically (Figure 4A) and Epstein–Barr virus-encoded small RNAs (EBER) by in situ hybridization (Figure 4B). In addition, CD99 and TTF-1 were positive immunohistochemically as in the tru-cut needle biopsy. CD3, CD5, CD10, cyclin D1, TdT, bcl-2, chromogranin A, synaptophysin, pancytokeratin, S100, SMA, desmin immunos-

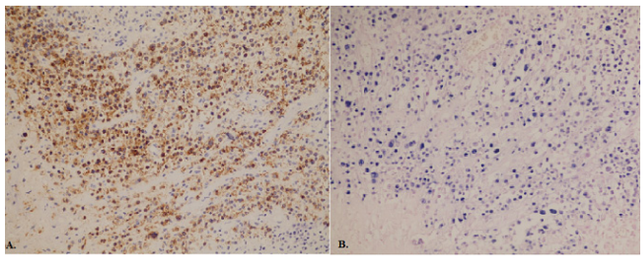

Figure 4. The immunopositivity for EBV in the tumor cells (Avidin-biotin-perxidase method, x100)(A). EBER in situ hibridization positivity in the tumor cells (x100)(B).

tains were negative.

Then, the tumor was diagnosed as chronic inflammation-related diffuse large B-cell lymphoma. The lymphoma infiltration was detected also in the wedge resection of the spleen with a tumor free margin. A bone marrow biopsy was performed by the Department of Medical Oncology and no tumoral involvement was found histopathologically. The patient received six cycles of adjuvant chemotherapy followed by radiotherapy in the Department of Medical Oncology. The patient was under follow-up without any relapse or complication for 2 years.

Discussion

Lymphomas compose of less than 5% of all primary chest wall malignancy [1]. Lymphomas that develop on the ground of pleural effusion or pyothorax have been reported especially among Japanese people. Chronic antigenic stimulation is thought to have an important role in neoplastic lymphoid transformation. Pyothorax, long-term use of antituberculosis drugs and antibiotics, extensive exposure to viral or bacterial subproducts and long-term radiation exposure due to diagnostic procedures may exist in the medical history of the chronic inflammation related diffuse large B cell lymphomas. In addition, it has been reported that p53 gene mutation has been detected in 67% of those patients with chronic inflammation related diffuse large B cell lymphomas [3-5].

In the literature, it has been reported that lymphoma develops 20 to 60 years after pleural inflammation [3]. In our case, the lymphoma developed on the base of surgical incision scar approximately 2 years after the pleural decortication. To the best of our knowledge, chronic inflammation-related lymphoma developed within such a short time as two years is the first case in the literature .

Histopathologic grading and prompt staging of the tumor is vital for the management and the prognosis. Previous studies proved the presence of EBER with in situ hybridization in tumor cells as an etiologic agent for lymphoma [5]. Our case also showed EBV positivity immunohistochemically. These findings supported the diagnosis of chronic inflammation-related lymphoma due to EBV in concordance with the literature. The positivity for TTF-1 (a marker indicating mostly the lung origin) of the tumor cells were consistently found both in the tru-cut needle biopsy and excision specimen that caused difficulty in the differential diagnosis. To the best of our knowledge this is the first lymphoma that showed TTF-1 immunopositivity, most probably as a cross-reaction.

Chronic inflammation-related diffuse large B-cell lymphoma has generally a worse prognosis. Five-year survival rate is about 20-30%. If remission with chemotherapy is enabled, overall survival rate of 50% can be obtained [3]. Total removal of the mass together with adjuvant chemotherapy has been reported to give better outcome. A study from Japan implied that extensive surgical resection produce five-year survival rate of 85.7% in case of lymphoma developed after empyema [6].

In our case, preoperative tru-cut needle biopsy result was compatible with small blue round cell tumor indicating Ewing/PNET or a tumor originated from lung. The left chest wall uptake belonging to a soft tissue lesion with a SUVmax value of 5.5 were seen on FDG-PET/CT images. In the light of all these findings, the case was considered as a primary thorax wall malignancy, and then the patient was operated. Postoperative pathology result of the wide excision of the mass in the chest wall was reported as chronic inflammation-related diffuse large B-cell lymphoma. According to the pathology result, the patient received adjuvant chemoradiation therapy and was under follow-up for two years without any complication.

It is well known fact that principal treatment modality for lymphomas are chemotherapy ± radiotherapy[6], however it was thought that adjuvant chemoradiation therapy can improve the prognosis in particular cases of tumors which have isolated chest wall involvement without definitive diagnosis at the beginning but with pathological diagnosis of lymphoma after total resection as in our case .

Competing interests

The authors declare that they have no competing interests.

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