

Case Report

Secondary leiomyosarcoma after treatment of diffuse large B cell lymphoma in a patient: a case report and literature review

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Abstract: Leiomyosarcoma (LMS) is a rare malignant tumor originating from the smooth muscle. To the best of our knowledge, there have been few reports about leiomyosarcoma as a second tumor of lymphoma. In the present report, a case of leiomyosarcoma in a 70-year-old Chinese man is discussed. This patient was diagnosed with diffuse large B cell lymphoma (DLBCL) and accepted chemotherapy seven years ago. The patient suffered from shortness of breath and pain in the right lower limb. Chest CT imaging showed multiple quasi-circular high-density nodules, comparable to that of a bullet wound. There was also an echo-clump in the middle segment of the right thigh by B ultrasonography. CT-guided percutaneous needle lung biopsy of the patient revealed leiomyosarcoma. The following B-ultrasound-guided needle aspiration biopsy of the right middle thigh mass showed spindle cell sarcoma, also revealed leiomyosarcoma. It is certain that secondary leiomyosarcoma occurred after treatment of diffuse large B cell lymphoma in this patient. Through this case, the risk of secondary tumor and reassessment of disease are discussed. Especially after a long time, this is important to avoid misdiagnosis.

keywords: Leiomyosarcoma, diffuse large B cell lymphoma (DLBCL), the secondary tumor

Introduction

There has been great progress in the diagnosis and treatment of malignant lymphoma in the past years. Most patients with conventional comprehensive treatment have long-term survival. However, the risk of secondary tumor is higher. Moser EC et al. reported that 4% of NHL patients died of second cancer [1]. Secondary tumors after treatment in long-term surviving lymphoma patients usually occurs as leukemia/MDS [1-3]. Breast cancer in young women with Hodgkin's disease has also been reported [4, 5]. But leiomyosarcoma as the second tumor of lymphoma is quite rare. In this report, a case of leiomyosarcoma is discussed involving a patient who was diagnosed with DLBCL and accepted chemotherapy seven years ago.

Case report

A 70-year-old man was diagnosed diffuse large B cell lymphoma (DLBCL) seven years ago. At that time, there were masses in his neck and

inguinal regions. The maximum diameter of the masses was about 6 cm. The patient had no fever, night sweats, or weight loss. The pathological result of lymph node was DLBCL, and the immunohistochemical staining was: CK(-), LCA(+), CD3(-), CD45RO(-), CD79a(+), Bcl-2(-), CD5(-), CyclinD1(-), CD20 foci(+). The histopathological image was not available because of time. At that time, the patient's chest CT scan was near normal (**Figure 1**). After 3 cycles CHOP regimen chemotherapy, he was in a stable condition and stopped the follow-up chemotherapy and clinic. Seven years later, he suffered shortness of breath and pain in the right lower limb. Through chest CT scan it showed that his lung was full of multiple quasi-circular high-density nodules, the longest diameter of which was about 3.9 cm (**Figure 2**). A mixed echo-clump in the middle segment of the right thigh was found by B ultrasonography, with a range of 99×34 mm, which was wrapped around the superficial femoral artery and the superficial femoral vein. Initially, the patient's DLBCL disease relapsed

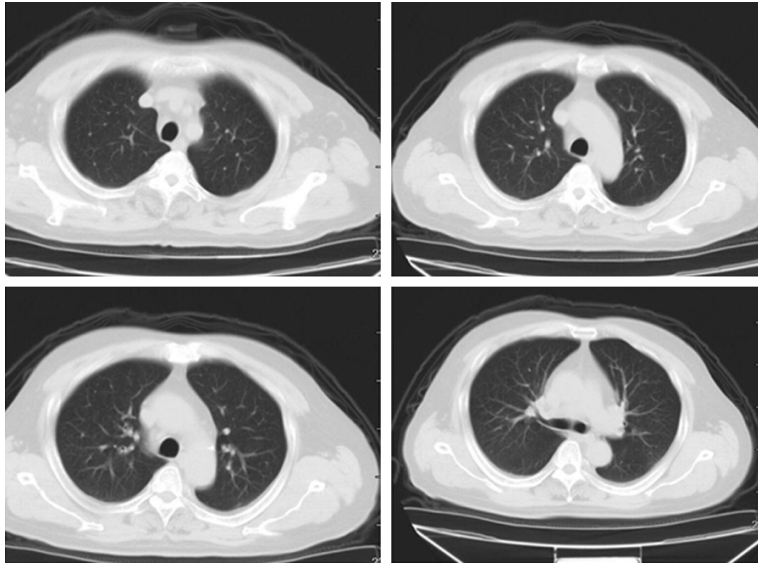


Figure 1. CT images of the patient's chest. There was no sign of tumor infiltration.

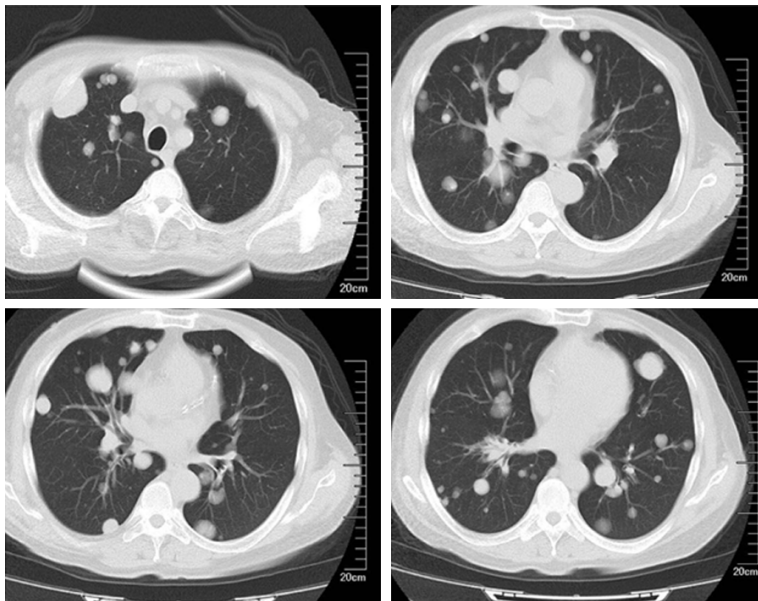


Figure 2. CT images of the patient's chest, There was multiple quasi-circular high-density nodules, the diameter of which was about 3.9 cm.

so he accepted two cycles treatment of CHOP regimen. The patient's discomfort was relieved and the mass in the middle segment of the right thigh was narrowed, but no significant change was observed regarding the pulmonary nodules in chest CT. CT-guided percutaneous needle lung biopsy of the patient revealed leiomyosarcoma. The heterogeneity of the cells was obvious, and the immunohistochemical

staining was: Vim(+), SMA(+), CK(-), TTF(-), CK5/6(-), NapsinA(-), CD20(-), CD3(-), CD34(-), S100(-), HMB45(-), CR(-), CD68(-), Ki67 (+30%) (**Figure 3**). The following B-ultrasound-guided puncture biopsy of the middle segment of the right thigh showed that spindle cell (**Figure 4**). Therefore, this patient was diagnosed with leiomyosarcoma.

Discussion

Leiomyosarcoma (LMS) is a rare malignant tumor originating from the smooth muscle, and constitutes 3-7% of all soft tissue sarcomas [6, 7]. The 5-year survival rate for LMS is 40%, but decreases to 10%-15% for high-grade LMS [8]. The most common is retroperitoneal type by location, followed by peripheral soft tissue (common in lower limbs) and blood vessels. LMS's prognostic factor is the location and size of the tumor. Local recurrence and distant metastasis commonly occur while lymph node metastasis is rare. The retroperitoneal type is easily transferred to the liver, while the other can be transferred to the lung. The case we report is the type of peripheral soft tissue. The tumor was transferred to the lungs through the blood flow. The standard of care for LMS is surgical resection when possible [9]. In the case of advanced disease, some sys-

temic agents have been considered active, historically doxorubicin and ifosfamide, and more recently gemcitabine-based combination, trabectedin, pazopanib, and eribulin [10]. So the patient had responses to CHOP regimen drugs which included doxorubicin and ifosfamide. In our case LMS was presented as the second tumor after treatment for DLBCL. The risk factor between LMS and lymphoma was exam-

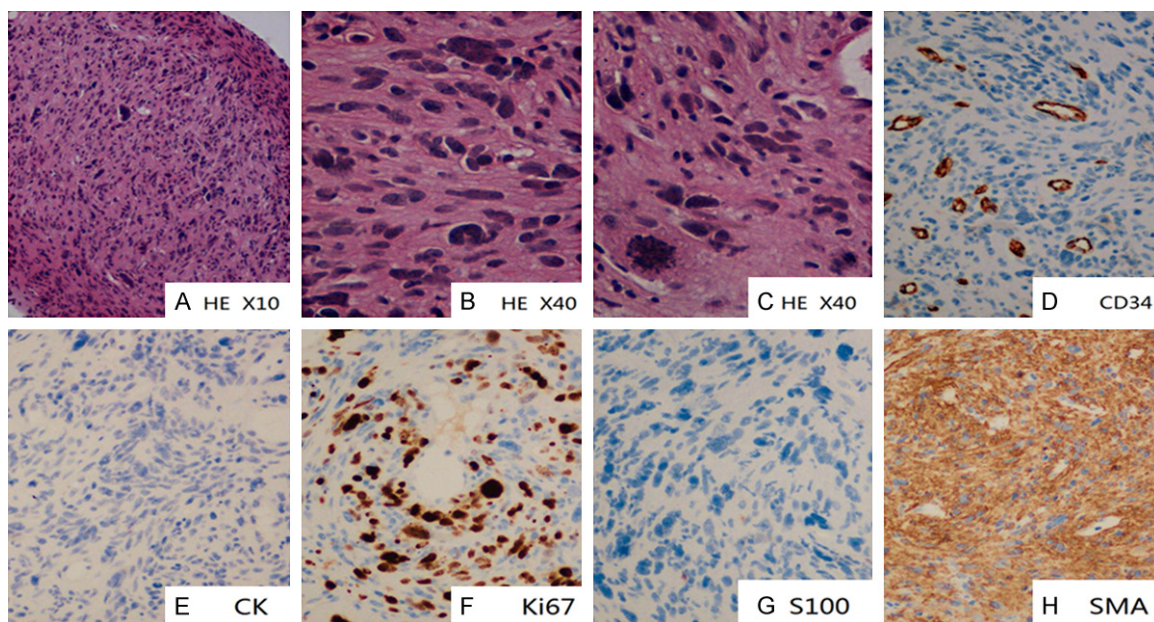


Figure 3. Pathological images of the patient' lung tissue. The heterogeneity of the cells was obvious and immuno-histochemical staining showed: CD34(-), CK(-), Ki67 (+30%), S100(-), SMA(+).

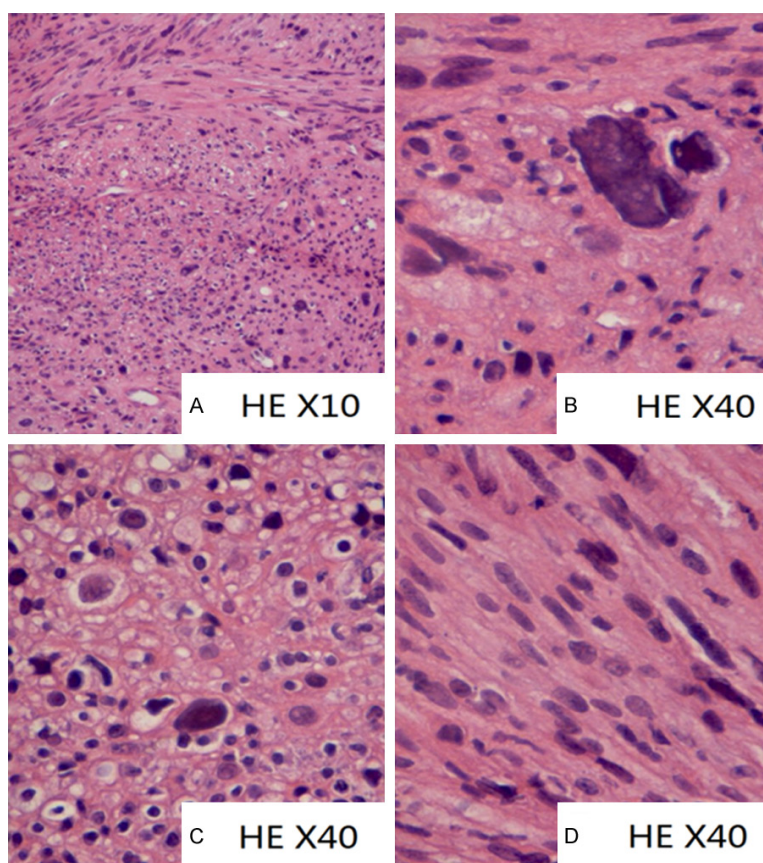


Figure 4. Pathological images of the patient' segment of the right thigh showing spindle cells.

ined. Irradiation, alkylating agent chemotherapy, and anti-metabolite use have been identified as risk factors that potentiate second malignancies [11, 12]. The monoclonal antibody rituximab has also been previously linked to second malignancies [13, 14]. But we review the treatment of the patient about DLBCL. He only accepted 3 cycles CHOP regiment chemotherapy, in which there was no radiotherapy, no rituximab and no anti-metabolite drug. Therefore, chemotherapy drug perhaps wasn't the major risk factor of the second cancer for this patient. Are the diseases of lymphoma and LMS have the same pathogenic factor? The following literature supports our hypothesis. There was a case report that a coincidence of subcutaneous leiomyosarcoma metastasis to same lymph nodes that were involved with lymphoma [15]. Simard EP et al. reported

that people diagnosed with AIDS during childhood remain at elevated risk for KS, NHL, and LMS [16]. There are reports that Epstein-Barr virus is associated with a wide spectrum of lymphomas [17, 18], and soft tissue sarcomas [19, 20]. So immunosuppression and Epstein-Barr virus are the same risk factor of LMS and lymphoma. Through this case we learn that the reassessment of disease is very important, especially after a long time, to avoid misdiagnosis.

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Disclosure of conflict of interest

None.

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