International Journal of Medical Science and Clinical Invention 5(10): 4101-4103, 2018
DOI:10.18535/ijmsci/v5i10.05 **ICV 2016: 77.2**
e-ISSN:2348-991X, p-ISSN: 2454-9576

Case Report

A Rare Primary Skeletal Lymphoma in the Tibia Mimicking Osteomyelitis

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Abstract:
The primary skeletal lymphoma is a rare disease that accounts for only 0.5% of all extra nodal, The pathogenesis of the lymphoma that occurs in the muscles is not clear. It has been suggested that the muscle lymphoma may originate from abnormal lymph nodes within the muscles. In this report , there is a case about this rare non-hodgkin lymphoma. A 42-year-old female patient presented to our clinic with complaints of pain in the left leg. In the bone biopsy from the lesion of the patient; it is seen that atypical cells with diffuse infiltration into adjacent striated muscle tissue The findings were evaluated as diffuse large B-cell lymphoma. Primary extra nodal lymphomas in the skeletal muscle are radiologically similar to osteomyelitis, so in the case of a patient complaining of pain and swelling in the adjacent skeletal muscle, primary extra nodal lymphoma should be considered in differential diagnosis, although it is rare

Keywords: lymphoma,skeletal,osteomyelitiw

Introduction

In malign hematologic diseases, secondary muscle infiltration from adjacent bone or lymph nodes is common. However, the development of malignant lymphoma in the skeletal muscle system is very rare. The primary skeletal lymphoma is a rare disease that accounts for only 0.5% of all extra nodal lymphomas, mainly (> 95%) B-cell and non-Hodgkin type lymphoma (NHL). Muscles affected most commonly; limbs, pelvis, and gluteal region , metastatic hematogenous or lymphatic spread or adjacent lymph nodes or adjacent extension of the bone (1). Skeletal lymphoma tends to occur in the lower limbs[6]. The pathogenesis of the lymphoma that occurs in the muscles is not clear. It has been suggested that the muscle lymphoma may originate from abnormal lymph nodes within the muscles[5]. In our article, there is a case report about this rare non-hodgkin lymphoma.

Case Presentation

A 42-year-old female patient presented to our clinic with complaints of pain in the left leg. She has no known co-morbidities. In laboratory analysis, it has been revealed that complete blood count; Hgb: 13,2 g/dl, Htc: 40,6%, leukocyte: 9380/mm^3 (77% neutrophil, 15,3% lymphocyte, 6% monocyte, 0.6% eosinophil) and platelet: 445000/mm^3. Liver function tests, renal function tests and serum electrolytes were within normal limits. Serum lactate dehydrogenase level was 216 U/L (normal range: 135-214) and beta-2 microglobulin level was 1.7 mg/L (normal range: 0.8-2.45).

Left-sided knee Magnetic Resonance imaging of the patient (20.07.2016); The edematous signal increases attracts attention in diafizer and proximal metaphyseal sections in left middle tibia and in the left fibula and representing contrast enhancements in these areas after IVKM.In the area described in the middle part of the left tibia, the cortex of the anteromedial face seems to be defective appearance. In the medial and lateral sections of the gastrocnemius muscle, an area of 9x8 cm between the muscle plans; it has been detected fluid collections, intermittent edematous signal increases and contrast enhancements. Findings may be significant in terms of osteomyelitis and deep tissue infections. Thorax CT (12.08,2016); peripherally located nodular density increments of less than 1 cm were observed in the middle lobe, right major fissure and lower lobe (pulmonary lymphoma?).

In the bone biopsy from the lesion of the patient; it is seen that atypical cells with diffuse infiltration into adjacent striated muscle tissue. Immunohistochemical studies were performed to detect cell structure, and tumor cells were found to be CK-PAN, Desmin, CD99, S-100, HMB-45, myoglobin and CD34 negative, CD45, vimentin CD79 a, CD20 positive. Some reactive lymphocytes with CD3 were found to be positive. The findings were evaluated as diffuse large B-cell lymphoma (due to CD79a and CD20 positivity).

In the PET-CT imaging in terms of tumor spread, pathologic F-18 FDG uptake was detected in the primer lesion covering the left leg muscle, in soft tissue lesion adjacent to right 11th rib on the back wall of the abdomen, in nodules of right lung lower lobe superior segment and in In cardiac nodules.

The patient underwent a protocol of 6 cycles of R-CHOP (rituximab375 mg/m^2, doxorubicin 50 mg/m^2, cyclophosphamide 750 mg/m2, vincristine 1.4 mg/m^2 and prednisone 100 mg).

On PET-CT after chemotherapy: Hyperdense area of 90.5x18.3 mm proximal to the posterior tibial muscle on the lateral aspect of the left lower limb has increased metabolic

activity (SUV max:3,4).In comparison with the patient's study dated 10/08/2016; treatment response is available with reduction in the number, size and activity of the metabolic active foci in the proximal left leg and the disappearance of the right lung nodules. Metastatic active distant organ metastasis associated with primary lesion was not detected.

Later on, radiotherapy was planned for the involved lesion of the patient. Between 31/01/2017-27/02/2017, With RAPIDARC device, A total of 40 GYexternal radiotherapy was applied to the left lower limb. Complications did not develop after radiotherapy. PET CT taken at 3 months after radiotherapy did not show any pathological activity. She was accepted in remission.

Figure 1: CD79a and CD20 positively stained lymphoma cells in the striated muscle tissue

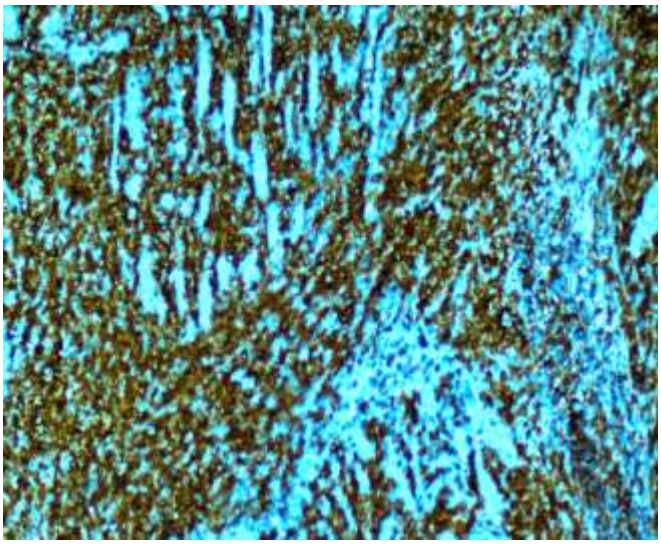
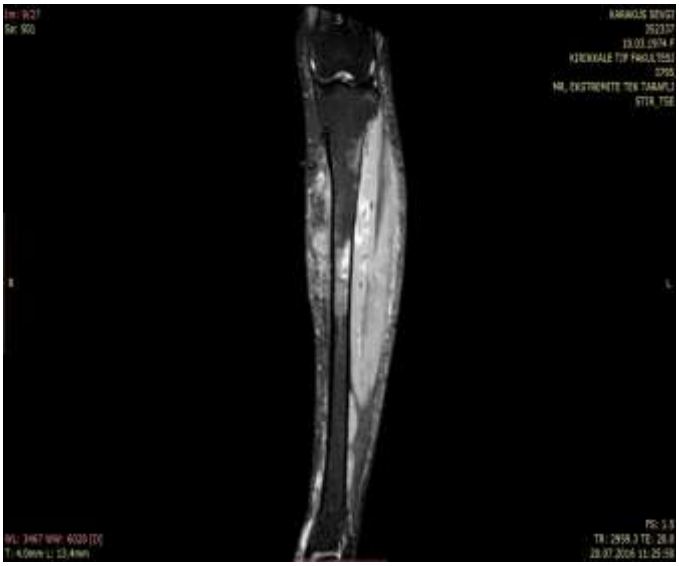


Figure2: Coronal contrast agent–enhanced fat-suppressed SPGR T1-weighted MR image shows heterogeneous enhancement in the muscle.



Discussion

Approximately 50% of non-Hodgkin lymphoma patients develop extra nodal involvement (secondary extra nodal involvement), whereas 10 to 35% of patients initially have primary extra nodal lymphoma[7]. Some of the extra nodal lymphomas develop from soft tissues, which account for less than 2% of soft tissue tumors. Non-Hodgkin's lymphoma (NHL) can develop in any extra nodal region including lymphoid tissue, but development in the soft tissue, including the skeletal muscle system, is very rare$_4$. Skeletal lymphomas constitute 0.5% of all extra nodal lymphomas. Two types of skeletal lymphoma have been identified: first, primary extra nodal intramuscular lymphoma and second, lymphoma characterized by intramuscular development of disseminated disease despite appearance in the muscular system[8].

B-cell lymphoma is the most common type of skeletal lymphoma; NK-cell lymphoma, T-cell lymphoma and plasmacytoma may also occur in the skeletal muscle system[8]. Clinically, these lymphomas usually manifest with soft tissue mass, swelling, and pain[9]. Most common histology Diffuse Large B-cell lymphoma (DLBCL), but almost all NHL strains have been described[10]. Hodgkin's lymphoma with soft tissue involvement is very rare[11]. 95% of the skeletal lymphomas are located in the extremities, the majority of which are localized in the lower extremities. Our case is also located in the lower extremity according to the literature.

The development of lymphoma primer in extra nodal regions can be explained by chronic inflammation, autoimmune process hypothesis; like primary thyroid lymphoma developing under autoimmune thyroiditis and intestinal lymphoma developing under celiac disease. It has been suggested that skeletal lymphoma may be associated with traumatic inflammation. Muscular lymphomas, such as those seen in drug injection sites or in the rectum of homosexual men or those seen in leg traumas are examples. Nevertheless, there is no leg trauma story in our patient.

The treatment of primary skeletal lymphoma is predominantly due to type of lymphoma. The prognosis of the primary skeletal lymphoma is highly poor when compared with lymph node lymphoma, especially in stage III-IV. For this reason, it is essential to choose the most effective treatment regimen. A DLBCL case with R-CHOP, the standard treatment, has been reported in this study[12]. The standard treatment for DLBCL is a combination of chemotherapy and immunotherapy known as R-CHOP (rituximab, doxorubicin, cyclophosphamide, vincristine and prednisone) with or without radiotherapy[13]. The combination of chemotherapy and radiotherapy has been reported to significantly increase disease-free survival and overall survival (OS) rates[14]. In addition, local radiotherapy following chemotherapy improves event-free survival (EFS) outcomes compared with chemotherapy alone[15].

Conclusion

Primer extra nodal lymphomas in the skeletal muscle are radiologically similar to osteomyelitis, so in the case of a patient complaining of pain and swelling in the adjacent skeletal muscle, primary extra nodal lymphoma should be considered in differential diagnosis, although it is rare.

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