

Ultrasound and Contrast-enhanced Ultrasound Patterns of Primary Psoas Major Muscle Diffuse Large B-cell Lymphoma: A Case Report

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Abstract

Diffuse large B-cell lymphoma involving skeletal muscle is rare and accounts for less than 0.1%, most frequently affecting the lower extremities. A 63-year-old man complained of a persistent fever, accompanied by waist discomfort. Computed tomography and ultrasound detected an ill-defined solid mass in the right psoas major muscle. Contrast-enhanced ultrasound showed that peripheral inhomogeneous hyper-enhancement with nonenhancing central necrotic area was seen in the mass. Then, ultrasound-guided biopsy was performed and revealed diffuse large B-cell lymphoma. This is the first case report demonstrated the primary psoas major muscle lymphoma and analyzed the ultrasound and contrast-enhanced ultrasound features of the tumor.

Keywords: Contrast-enhanced ultrasound; diffuse large B-cell lymphoma; primary skeletal muscle lymphoma; psoas major muscle; ultrasound.

Introduction

Primary extranodal NHL is a type of non-lymphoma Hodgkin's (NHL) that develops in tissue other than the lymph nodes, Waldeyer's ring, thymus and spleen [1, 2]. Diffuse large B cell lymphoma (DLBCL) is the most frequent kind of NHL, particularly among the elderly [3], and has a subtype with extranodal presentation [4]. Extranodal DLBCL with primary skeletal muscle involvement is extremely unusual, especially in the thigh and calf areas [5]. Primary involvement of psoas major muscle is quite uncommon and only few cases were mentioned in the literature [6]. Some radiological modalities such as ultrasound (US), magnetic resonance imaging (MRI), computed tomography (CT) and positron emission tomography/computed tomography (PET/CT) have been utilized to diagnose skeletal muscle lymphoma. Contrast-enhanced ultrasound (CEUS) also plays an important role in the evaluation of skeletal muscle lymphoma. However, a biopsy is necessary to make definite diagnosis. This case report mainly analyzed the US and CEUS features of DLBCL with primary involvement of psoas major muscle.

Case presentation

A 63-year-old male patient was admitted to local hospital due

to a fever and an absence of an inciting event prior to onset 1 month ago, with a body temperature of 37.3 °C to 37.4 °C, accompanied by fatigue and poor diet. The blood routine examination prompted leukopenia and thrombocytopenia. The patient received systemic therapy and the symptoms were relieved. After discharge, he had orally taken drugs for leucopenia. When the symptoms reappeared, the patient returned to the local hospital 6 days ago, mostly at night, with a maximum body temperature of more than 38°C, accompanied by fatigue and waist discomfort. Finally, the patient was admitted to the Department of Hematology in our hospital with "thrombocytopenia".

The blood test results were as follows: White cell count, $3.0 \times 10^9/L$; hemoglobin, 103g/L; platelets, $84 \times 10^9/L$, which were lower than the reference value. Bone marrow biopsy revealed non-Hodgkin large B-cell lymphoma, predisposing to diffuse large B-cell lymphoma.

CT scan revealed a mixed density mass in the right psoas muscle, with indistinct margins. The CT value was about 24HU and exudative changes were seen around both kidneys. But the size and shape of both kidneys were normal (**Figure 1**). US examination of right waist revealed large ill-defined, irregular hypoechoic solid mass of 34mm × 27mm within the back of

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the right kidney (**Figure 2A**). Color Doppler US showed stellate blood flow signal inside the mass (**Figure 2B**). One week later, on CEUS, the hypoechoic lesion in the right psoas major muscle showed inhomogeneous hyper-enhancement at 10s after injection of Sonovue, with irregular nonenhancing regions in the lesion (**Figure 3A-B**), and washout compared with surrounding tissue in the late phase (**Figure 3C**). CEUS illustrated a solid hypervascular mass with formation of central necrotic area in the right psoas muscle.

Therefore, US-guided biopsy was performed (**Figure 3D**) and revealed diffuse large B-cell lymphoma (germinal center B-cell type). Immunohistochemical examination results showed CD20 (+), Mum-1 (+), CD10 (+), C-MYC (weak +, about 50%), Bcl-6 (+), EMA (-), CK (-), S-100 (-), Bcl-2 (-), CD34 (-), SALL4 (-), CK8/18 (-), Desmin (-), CD3 (-), Ki67 (about 80%). In situ hybridization results showed EBER (-) (**Figure 4**). Finally, the malignancy was categorized as stage IV B, IPI 4 high-risk group according to the Ann Arbor system. The R-CHOP (rituximab, cyclophosphamide, doxorubicin, vincristine and prednisolone) chemotherapy regimen was used on the patient. However, on the first day, during the instillation of rituximab, the patient presented a fever with chills, and the body temperature up to 40°C. Rituximab was stopped immediately, and the desensitization treatment was performed. The CHOP (cyclophosphamide, doxorubicin, vincristine and prednisolone) chemotherapy was started the next day. Till this moment, after the first chemotherapy of the patient, CT scan showed that the mass at the right psoas muscle was significantly reduced (**Figure 5**).



Figure 1: Computed tomography image demonstrated a vague margin and mixed density mass in the right psoas muscle (arrows).

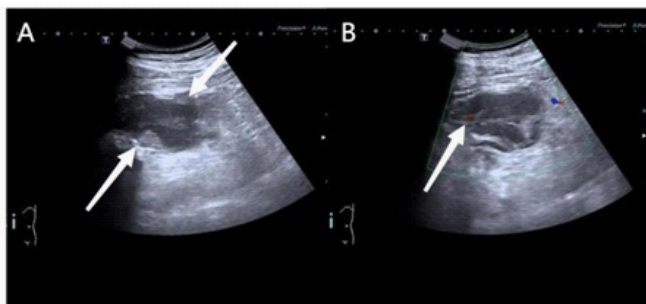


Figure 2: Illustrations of ultrasound hypoechoic lesion in the right waist. (A) B-mode ultrasound revealed a large poorly defined hypoechoic mass (arrows). (B) Color Doppler ultrasound showed stellate blood flow signal inside the mass (arrow).

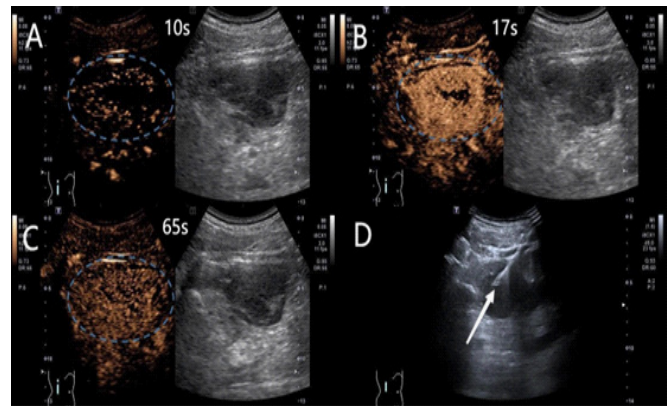


Figure 3: Illustrations of contrast-enhanced ultrasound hypoechoic lesion in right psoas major muscle. (A) The contrast agent started to enter the lesion 10 s after Sonovue injection. (B) The lesion showed inhomogeneous hyper-enhancement compared with surrounding psoas major muscle at 17 s, with irregular nonenhancing regions in the lesion. (C) Mild washout was seen at 65 s. (D) Ultrasound-guided core needle biopsy was performed subsequently (arrow).

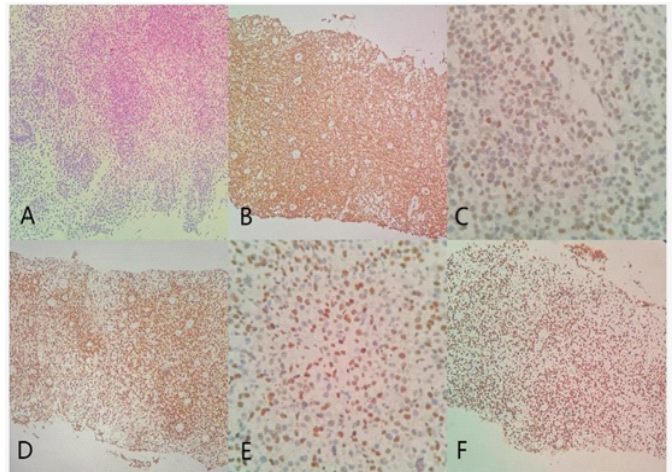


Figure 4: Pathological and immunohistochemical examination findings. (A; 100×) Hematoxylin-eosin staining showed medium sized tumor cells. (B–F) Immunohistochemical staining showed the tumor cells were positive for CD20 (B; 100×), Bcl-6 (C; 200×), Mum-1 (D; 100×) and weak positive for C-MYC (E; 200×). Ki-67 proliferation index was 80% (F; 100×).



Figure 5: Computed tomography image demonstrated the right psoas muscle mass was significantly reduced after the first chemotherapy (arrows).

Discussion

Extranodal NHL comprises for 20–30% of all cases, and primary muscular lymphoma is an uncommon form that typically affects the lower limbs [7]. Only 8 lymphoma cases out of 7,000 (0.1%) were diagnosed as muscle lymphoma of the extremi-

ties, according to a 10-years study by Travis et al [8]. Diffuse big B-cell immunophenotype primary skeletal muscle NHL is extremely uncommon which was first reported by Kandel et al [9] in 1984 and is found in about 0.5% of extranodal lymphomas. Therefore, only a few cases of DLBCL developing largely from skeletal muscle, particularly the psoas major muscle, have been reported. It is a significant item to consider in the differential diagnosis of other cancers, despite its rarity.

Several research have reported on imaging modalities for the diagnosis of skeletal muscle lymphoma. Using CT, the tumors usually appear as low density or iso-density masses in the skeletal muscle [10]. Although nonspecific, US is useful and convenient for delineating soft tissue lesions. The sonographic, as an initial investigation tool of this lesion, showed an ill-defined hypoechoic mass with preservation of the underlying muscle architecture. Skeletal muscle lymphoma usually takes the form of irregular, poorly defined hypoechoic masses with a texture similar to muscular fibers that maintain continuity with adjacent muscles [11]. Other studies reported that the color Doppler US may show hypervascularity within the lymphoma which is similar to our report [5, 12].

Musculoskeletal soft tissue tumors are very common and diverse lesions, ranging from benign and malignant tumors to tumor-like lesions [13]. Vascular supply is an important identification point to differentiate benign from malignant tumors [14]. The non-hepatic CEUS, an emerging application, is concerned with the use of microbubble ultrasound contrast outside the liver which is a recognized imaging tool in the characterization of lesions to increase signal backscattering from the blood [15]. CEUS can detect both tumor angiogenesis and vascularity. Loizides et al [16] identified peripheral enhancement with a non-enhanced central area have a positive predictive value (PPV) of 86 percent and a negative predictive value (NPV) of 88 percent in discriminating benign from malignant tumors and the perfusion pattern is consistent with our results. Another study showed that the rapid arterial vascularization time (<11 s) was linked to a higher risk of malignancy [17] and the arterial perfusion in our case was just very rapid (10 s).

However, only a few earlier reports of CEUS used in skeletal muscle lymphoma have been reported, and the relationship between CEUS and pathological characteristics remains unclear. In a study about CEUS and intestinal lymphoma reported that high proportion of cases had arterial hyperechoic enhancement followed by venous wash out, as well as a high rate of central necrosis (61.1%) was observed which was more frequent in aggressive subtypes [18]. A study by Trenker et al [19] showed that renal lymphoma consistently showed wash-out in the late phase, and it can help to distinguish renal lymphoma from benign tumors. Therefore, the above studies confirmed that the extranodal lymphoma is a rich blood supply mass which grows rapidly, and it is prone to central necrosis. Thus, our results showed that skeletal muscle DLBCL displayed almost identical CEUS patterns.

In conclusion, DLBCL with primary involvement of psoas major muscle is rare, and both US and CEUS are advantageous tools that assist in diagnosing the disease. Involvement of inhomogeneous hyper-enhancement at the early stage with formation of no enhancement central necrotic area and phenomenon of washout in the late phase are symptomatic of primary skeletal

muscle lymphoma on CEUS. US-guided core needle biopsy is still a safe and practical method for obtaining sufficient tumor materials to make a definitive diagnosis [20].

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Conflict of Interest: The authors declare no conflict of interest.

Data Availability Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

Declarations: Author's institution does not require ethical approval for publication of a single case report. Written informed consent was obtained from the patient for publication of this case report and the accompanying images.

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