CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 7 (2015) 161–164

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

# Extranodal lymphoma originating in the gluteal muscle with adjacent bone involvement and mimicking a soft tissue sarcoma

Morihiro Katsura [a,*], Hirokazu Nishina [b], Yasushi Shigemori [c], Takaya Nakanishi [d]

[a] Department of Surgery, Okinawa Prefectural Hokubu Hospital, Okinawa, Japan
[b] Department of Internal Medicine, Okinawa Prefectural Hokubu Hospital, Okinawa, Japan
[c] Department of Radiology, Okinawa Prefectural Hokubu Hospital, Okinawa, Japan
[d] Department of Pathology and Cell Biology, University of the Ryukyus, Graduate School of Medicine, Okinawa, Japan

ARTICLE INFO

*Article history:*
Received 17 October 2014
Received in revised form 12 January 2015
Accepted 13 January 2015
Available online 15 January 2015

*Keyword:*
Gluteal mass
Extranodal lymphoma
Diffuse large B cell lymphoma
PET

ABSTRACT

*INTRODUCTION:* Extranodal lymphoma (ENL) in the muscles is a rare manifestation of non-Hodgkin lymphoma (NHL). The aim of this case report is to describe and evaluate the clinical presentation and important radiologic features of ENL affecting the musculoskeletal system.
*PRESENTATION OF CASE:* We present a 52-year-old female with a 3-week history of left gluteal pain. Computed tomography (CT) showed a non-uniformly early enhancing mass in the left gluteal muscle, the tumor demonstrating central necrosis and adjacent bone involvement. Fluorine-18 fluorodeoxyglucose positron emission tomography ($^{18}$F-FDG PET)/CT showed areas of increased $^{18}$F-FDG uptake in the left gluteal musculature, pelvic bones, para-aortic and mediastinal lymph nodes and both lungs. Histopathological examination showed a diffuse large B cell lymphoma (DLBCL). After 8 cycles of R-CHOP chemotherapy, the mass in the left gluteal muscle has completely disappeared
*DISCUSSION:* Although destructive tumor originating in the gluteal muscle with adjacent bone involvement is more common in soft tissue sarcoma, lymphoma should be regularly included in the differential diagnosis. While CT is a useful modality for assessing soft tissue masses, disruption and injury of the surrounding tissues, PET/CT fusion is superior for the detection of unexpected extranodal sites of disease, or for exclusion of disease in the presence of nonspecific extranodal CT findings.
*CONCLUSION:* A rapid growth pattern and destructive masses that invade adjacent structures on CT are key findings of DLBCL, and $^{18}$F-FDG PET/CT is a useful imaging modality to accurately determine the disease stage and disease aggressiveness of NHL.


## 1. Introduction

Malignant lymphomas are a heterogeneous group of malignancies of the B cells or T cells that usually originate in the lymph nodes, although they can originate in any part of the body [1–3]. However, extranodal lymphoma (ENL) in the muscles as an isolated manifestation is very rare, accounting for 1.5–8.3% of non-Hodgkin lymphoma (NHL) cases [4–6]. Clinically, the presence of cortical bone destruction in association with large soft-tissue masses arising in the skeletal muscle, favors the diagnosis of soft tissue sarcoma, malignant musculoskeletal tumor such as malignant fibrous histiocytoma (MFH), malignant neurogenic tumor (MPNST) or metastasis rather than malignant lymphoma [3,4,7,8].

Despite advances in imaging devices over the past decade, accurate diagnosis of soft tissue tumors remains a challenge for clinicians, requiring a close team approach between the surgical oncologist, musculoskeletal radiologist and pathologist. Here we present a rare case of extranodal lymphoma originating in the gluteal muscle, with aggressive bone erosion, indicative of primary bone involvement. We believe that our case adds an important piece of evidence to the clinically relevant problem of appropriate diagnostic strategy for an unusual presentation of malignant lymphoma.

## 2. Case report

A 52-year-old female with a 3-week history of progressive left gluteal pain was referred to our outpatient surgical department. She was previously healthy but had a history of night sweats and weight loss over the prior 2 months. Upon physical examination, a large firm mass was observed in the left buttock, with no inflammation

* Corresponding author at: Department of Surgery, Okinawa Prefectural Hokubu Hospital, 2-12-3, Onaka, Nago, Okinawa 905-8512 Japan. Tel.:+ 81 980 52 2719; fax: +81 980 54 3965.
*E-mail addresses:* morihiro@bj8.so-net.ne.jp (M. Katsura), carrot_and_whip_hiro@yahoo.co.jp (H. Nishina), shigemori-sin@umin.ac.jp (Y. Shigemori), h097509@med.u-ryukyu.ac.jp (T. Nakanishi).

http://dx.doi.org/10.1016/j.ijscr.2015.01.024

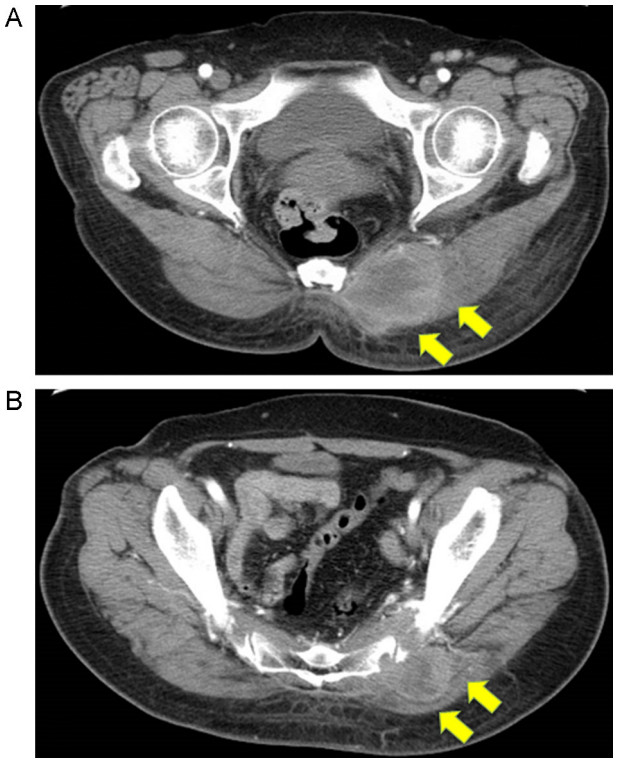


**Fig. 1.** Contrast-enhanced computed tomography (CT) of the pelvis demonstrating the left gluteal mass (arrow). (A) CT showed a non-uniformly enhancing hypervascular mass with central necrosis in the left gluteal muscle. (B) The tumor demonstrated adjacent bone involvement with destruction of the sacroiliac joint.

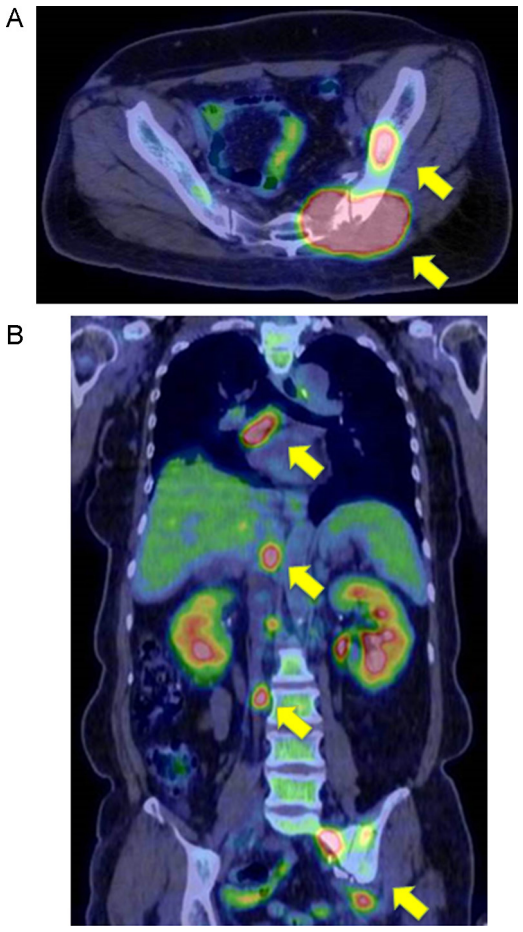


**Fig. 2.** Fluorine-18 fluorodeoxyglucose positron emission tomography ($^{18}$F-FDG PET)/CT images. (A) Axial $^{18}$F-FDG PET/CT view demonstrated large areas of increased $^{18}$F-FDG uptake in the left gluteal musculature [maximum standard uptake value ($SUV_{max}$) = 34], the posterior aspect of the left ileum (arrow) and the sacrum. (B) Whole-body $^{18}$F-FDG PET/CT identified strong $^{18}$F-FDG uptake by the para-aortic and mediastinal lymph nodes (arrow), and faint scattered $^{18}$F-FDG uptake by both lungs ($SUV_{max}$ = 3.8).

of the skin. No peripheral lymphadenopathy was detected despite thorough examination of the whole body. Standard biochemistry and hematology studies revealed normal results, except for mild elevation of serum lactic dehydrogenase (LDH) and C-reactive protein (CRP). Contrast-enhanced computed tomography (CT) scan of the pelvis revealed a non-uniformly early enhancing mass, approximately 51 × 64 mm in size, in the left gluteal muscle (Fig. 1A). The tumor demonstrated central necrosis and adjacent bone involvement, with destruction of the sacroiliac joint (Fig. 1B). CT scan of the chest showed patchy consolidation in the lower lobes of both lungs. Fluorine-18 fluorodeoxyglucose positron emission tomography ($^{18}$F-FDG PET)/CT demonstrated a large area of increased $^{18}$F-FDG uptake in the left gluteal musculature [maximum standard uptake value ($SUV_{max}$) = 34], the posterior aspect of the left ileum and the sacrum (Fig. 2A). Whole-body $^{18}$F-FDG PET/CT identified intense $^{18}$F-FDG uptake by the para-aortic and mediastinal lymph nodes, and faint scattered $^{18}$F-FDG uptake by both lungs (Fig. 2B). Based on these findings, the clinical and radiographic differential diagnosis was soft tissue sarcoma, malignant lymphoma or metastasis derived from small cell carcinoma of the lung. We then proceeded with an open biopsy of the left gluteal mass for further diagnosis and treatment planning. Pathological studies of the specimen demonstrated dense and diffuse infiltration and proliferation of large atypical lymphoid cells, accompanied by small lymphocytes (Fig. 3A). Immunohistochemical studies showed that the large atypical lymphoid cells were positive for LCA, CD20 and bcl-6, and were negative for cytokeratin, S-100, alpha-SMA and bcl-2 (Fig. 3B). Further, approximately 45% of the large atypical lymphoid cells were positive for MIB-1(Ki-67). Thus, from these results, we diagnosed diffuse large B cell lymphoma (DLBCL) of the left buttock. After staging work-up, including bone marrow biopsy, the patient was finally diagnosed with stage IV DLBCL, low-intermediate risk of the international prognostic index (IPI). Immediately after diagnosis, the patient has received 8 cycles of R-CHOP chemotherapy, and as a result the mass in the left gluteal muscle has completely disappeared (Fig. 4A, B). The patient achieved complete remission after chemotherapy and is currently under the regular follow-up evaluation.

## 3. Discussion

Malignant lymphomas in the musculoskeletal organs are predominantly a manifestation of disseminated lymphoma [3,4]. Common sites for the development of ENL are the skin, head, neck and gastrointestinal tract; primary lymphoma of the skeletal muscle is extremely rare [1–4]. To date, only a few cases of malignant lymphoma originating in the gluteal muscle have been reported in the English literature [5,7,9,10]. Textbooks present the common aspects of ENL in detail, but the published literature on the unexpected manifestations of NHL, such as soft tissue masses invading adjacent structures, is inadequate. These tumors can have an atypical presentation, which can lead to diagnostic difficulty.

The most meaningful radiologic features of ENL affecting the musculoskeletal system are permeative lytic destruction, although mixed lytic and blastic (sclerotic) lesions have been reported [3,11,12]. Patients with primitive or poorly differentiated tumors were more likely to have bone involvement than patients with well differentiated lymphomas [12]. Although the compartmental