Non-Hodgkin's Lymphoma with Intraspinal Involvement Mimics Bilateral Thoracolumbar Plexopath

Wei-Hao Lin, Meng-Ni Wu*

Abstract

Purpose: Non-Hodgkin lymphoma (NHL) is the most common type of lymphoma, and its extranodal manifestation is rare. Skeletal muscle involvement is noted in only 1.1% of patients with NHL. Here, we present a case of high-grade B-cell lymphoma (HGBL); it infiltrated the left neural foramina from the left psoas muscle before encroaching on the whole spinal canal and subsequently invading the contralateral neural foramina from T12 to L3.

Case report: A 43-year-old man with HGBL who could function independently presented with numbness and weakness of the left thigh 2 months after a diagnosis of infiltrative lymphoma in the left psoas muscle. His symptoms were urine incontinence and unsteady gait. A neurological examination revealed weakness in the left psoas and quadriceps with hyporeflexia and hypesthesia. Lumbar spine magnetic resonance imaging (MRI) revealed intraspinal extradural invasion from T12 to L3 with multiple left-sided root compression despite the resolution of primary psoas lymphoma. At 6 weeks after symptom onset, his symptoms progressed to weakness, numbness, and hyporeflexia of the bilateral lower extremities with preserved anal sensation. Follow- up MRI revealed the progression of intraspinal invasion, which spread through the spinal canal and invaded the contralateral neural foramina from T12 to L3. The patient was finally bound to a wheelchair.

Conclusion: Clinicians must check for possible intraspinal involvement in patients with HGBL, particularly patients with known paraspinal soft-tissue involvement. The resolved infiltration of the soft tissue does not preclude the possibility of further neurological involvement. Additionally, MRI may provide higher resolution findings for clarifying the structure of the neural foramina and thecal sac.

Keyword: Non-Hodgkin's Lymphoma, high-grade B-cell lymphoma, plexopathy.

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INTRODUCTION

Non-Hodgkin lymphoma (NHL) is the most common type of lymphoma and typically presents with generalized lymphadenopathy. Primary extranodal lymphoma is rare. The gastrointestinal tract or bone marrow is the most common site of extranodal lymphoma. The skeletal muscle is rarely involved – only 1.1% of NHL cases

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involve the musculoskeletal system—and the extremities, pelvis, and gluteal regions are the most commonly affected areas1. Here, we present a rare case of high- grade B-cell lymphoma (HGBL), a subtype of NHL, with infiltration of the left psoas muscle and spreading through the spinal canal with the invasion of the thecal sac, resulting in bilateral thoracolumbar plexopathy with an initial resolution of lymphoma.

CASE PRESENTATION

A 43-year-old man had HGBL invading the pancreatic head and encasing the common hepatic artery as well as a superior mesenteric artery with MYC and BCL2 expression. After six chemotherapy sessions with R-CHOP (rituximab, cyclophosphamide, doxorubicin hydrochloride, vincristine sulfate, and prednisolone), flank soreness developed on the left side. Abdominal computed tomography (CT) and magnetic resonance imaging (MRI) revealed recurrent B-cell lymphoma in the left psoas muscle with the invasion of the left neural foramina (Fig 1A and 1B). Another course of chemotherapy with R-ICE (Mabthera, Carboplatin, and etoposide with ifosfamide) was administered. Two months later, the patient presented with numbness and weakness of the left thigh with urine incontinence and unsteady gait. The patient denied any symptoms of upper extremities, saddle anesthesia, stool incontinence, or constipation. Neurological examination revealed weak left hip flexion and knee extension (the detailed muscle strength with muscle power scale is reported in Table 1), hyporeflexia of the left knee jerk, normal plantar reflex, and hypesthesia at the left side

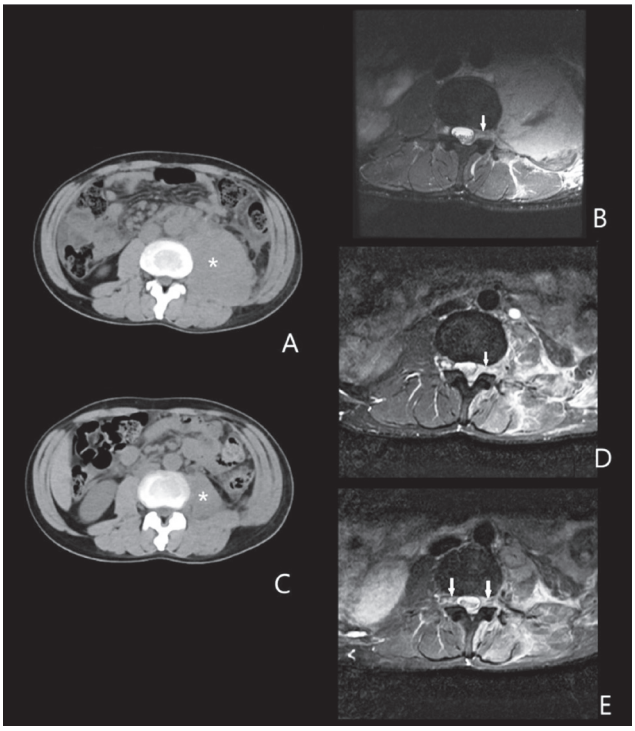

Figure 1. MRI revealed the progression of intraspinal involvement (arrow sign), although the resolution of the psoas tumor (star sign) was identified through abdominal CT. The tumor initially invaded the epidural space through the left intervertebral foramina; it then encased the whole spinal canal with the compression of the thecal sac and finally invaded the contralateral intervertebral foramina. All the images are at the L3 level, and all MRI sequences are T2-weighted images.
A. Abdominal CT performed 2 months before the presentation of symptoms; B. Lumbar spine MRI at 2 months before symptom onset; C. Abdominal CT within 1 week after symptom onset; D. Lumbar spine MRI at 3 weeks after symptom onset; E. Lumbar spine MRI at 6 weeks after symptom onset; Arrow sign: intraspinal invasion. Star sign: psoas muscle; Arrow sign: intraspinal involvement.

T12-L3 dermatome. Despite the partial resolution of infiltration in the psoas muscle, as observed on an abdominal CT (Fig 1C), lumbar MRI revealed intraspinal extradural invasion (Fig 1D).

After 6 weeks since the onset of the numbness and weakness of the left thigh, his symptoms progressed to weakness and numbness in the bilateral lower extremities, which made him unable to stand. Neurological examination revealed paraparesis with poor muscle power in the left lower limb (the detailed muscle strength score is listed in Table 2), hypesthesia below the T12 level on the left side and L3 on the right side, hyporeflexia of the bilateral knee jerk and ankle jerk, and no response of the left plantar reflex. Anal sensation was still present. Because of poor response to the prior chemotherapy regimen, a hyper-CVAD regimen (fractionated cyclophosphamide, vincristine, doxorubicin, and dexamethasone alternating with high-dose methotrexate and cytarabine) was administered for lymphoma. The nerve conduction velocity test revealed a decreased amplitude in the bilateral peroneal, tibial, and sural nerves with delayed F-wave and H-reflex latencies, suggesting bilateral lumbar plexopathy.

Electromyography demonstrated abnormal resting activity in the right vastus medialis, bilateral tibialis anterior, and left gluteus maximus; neurogenic motor unit action potentials in the left tibialis anterior on exertion; and a decreased recruitment pattern in the right vastus medialis, suggesting diffused neuropathic changes (i.e., bilateral plexopathy or peripheral neuropathy). Follow-up lumbar MRI did not indicate the progression of intraabdominal lymphoma or a mass. By contrast, the infiltration of lymphoma from the left psoas muscle spread through the left neural foramina, then encroached to the whole spinal canal with thecal sac compression, and finally invaded the right neural foramina of T12 to L3. (Fig 1E) The cerebrospinal fluid could not be collected. However, isolating and decompressing nerve roots are difficult after the tumor encroaches to the spinal cord, leading to bilateral multilevel foraminal invasion. The patient was finally bound to a wheelchair.

Table 1. The detailed muscle strength at the time point of the second MRI

MRC at the second MRI		
MRC on the right side	Muscle	MRC on the left side
5/5	Iliopsoas muscle	2/5
5/5	Quadriceps femoris	2/5
5/5	Tibialis anterior	5/5
5/5	Extensor hallucis longus	5/5
5/5	Gastrocnemius	5/5

Table 2. The detailed muscle strength at the time point of the third MRI

MRC at the third MRI		
MRC on the right side	Muscle	MRC on the left side
4/5	Iliopsoas muscle	1/5
4/5	Quadriceps femoris	1/5
2/5	Tibialis anterior	1/5
1/5	Extensor hallucis longus	1/5
1/5	Gastrocnemius	1/5

DISCUSSION

NHL is the most common type of lymphoma, and B-cell lymphoma accounts for most of the cases of NHL. HGBL, a newly established category in the 2016 World Health Organization (WHO) classification, replaces the previous category of B-cell lymphoma with indistinguishable features between diffuse large B-cell lymphoma and Burkitt lymphoma. HGBL with MYC and BCL2 expression is termed double-hit lymphoma, and the involvement of the central nervous system is frequently observed with relapse. Patients with HGBL have an aggressive clinical course and exhibit a poor response to standard chemotherapy regimens. Even high-dose chemotherapy or autologous stem-cell transplantation does not improve outcomes. The median overall survival is 1.5 years 2, 3.

The skeletal muscles are rarely involved in NHL (a phenomenon reported in only 1.1% of patients). Because of the rarity of muscle involvement in HGBL, related data are not available. In NHL, the extremities, pelvis, and gluteal regions are the most commonly affected muscles, and the psoas muscle is rarely involved1. Epidural spinal cord compression occurs in 0.1% to 6.5% of patients with NHL, and the thoracic spine is the most commonly affected (followed by the lumbar and cervical spine).

Patients with lymphoma involving the epidural space typically exhibit preceding paraspinal soft-tissue involvement and subsequent invasion into the epidural space through the intervertebral foramina without bone destruction, as observed in our case 4. To the best of our knowledge, although NHL usually invades the epidural space through the intervertebral foramina, the infiltration of NHL spreading through the spinal canal with the involvement of the bilateral intervertebral foramina has not been reported. In critically ill patients, paraparesis may mislead the clinician to consider it a neuropathy related to critical illness. Moreover, in patients with initial intraabdominal lymphoma, the asymmetric involvement of the bilateral lower legs may be misdiagnosed as bilateral plexopathy. We highlight the importance of determining the possible spinal canal involvement in patients with NHL with bilateral neurological deficits, particularly for those with known paraspinal soft-tissue involvement.

Furthermore, adjacent soft tissue or neurological involvement cannot be excluded based on the resolution of initial lymphoma.

Compared with CT, MRI provides better resolution for separating the normal tissue from the abnormal soft tissue5. In our case, abdominal CT revealed the resolution of mass in the left psoas muscle, whereas MRI revealed the epidural invasion of lymphoma from the intervertebral foramina. Therefore, we suggest MRI, instead of CT, for follow-up in patients with NHL with paraspinal soft-tissue involvement.

Surgery, radiotherapy, or a combination of both are therapeutic options for spinal cord compression by lymphoma. Surgical intervention can be simultaneously used for obtaining a tissue specimen and decompression. However, prior studies have reported no significant difference in the outcome between combination with decompressive laminectomy and radiotherapy and spinal radiation only. Typically, NHL is a chemo- sensitive tumor, and chemoradiotherapy may provide a favorable outcome, except for HGBL, which is a chemoresistant tumor6. Pretreatment neurologic status, especially motor function, is the most crucial prognostic factor for overall survival and functional outcome 7. Unfortunately, the patient was finally discharged without surgical intervention because of difficulty in isolating and decompressing nerve roots after the tumor encroaches the spinal cord with bilateral multilevel foraminal invasion.

In conclusion, we reported a rare case of HBGL where it infiltrated the left psoas muscle before spreading through the spinal canal and subsequently invading the contralateral intervertebral foramina from T12 to L3. Clinicians must identify any possible encroachment to the spinal canal in patients with HBGL, particularly patients with known paraspinal soft-tissue involvement. The resolved infiltration of the soft tissue cannot preclude the possibility of neurological involvement. Additionally, MRI may provide higher resolution findings for clarifying the structure of the neural foramina and thecal sac.

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