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SNI: Spine

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Case Report

Primary spinal Burkitt's lymphoma: Case report and literature review

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ABSTRACT

Background: Burkitt's lymphoma is a non-Hodgkin B-cell lymphoma, occurring mostly in Equatorial Africa. According to the WHO, classification is three different variants: sporadic, endemic, and immunodeficient-associated. Here, we present a patient with "sporadic" primary epidural Burkitt's lymphoma resulting in chronic low back pain (LBP).

Case Description: A 63-year-old female presented with a 2-month history of LBP and the left lower extremity sciatica. The thoracolumbar MRI showed a L5 irregular, osteolytic epidural lesion that was hypointense on T1-weighted images, hyperintense on STIR studies, and inhomogeneously enhanced with contrast. Additional hypointense lesions were also seen at the L2, L3, and L4 levels. The patient underwent a L4-L5 laminectomy for piecemeal epidural resection of tumor, and a L4-S1 transpedicular screws/rod fusion. In addition, a L2-L3 radiofrequency ablation was performed. The histological examination documented a primary "sporadic" spinal Burkitt's lymphoma. The patient subsequently was treated with both radiotherapy/chemoradiotherapy

Conclusion: Primary "sporadic" spinal Burkitt's lymphoma is rare. Following tumor resection, adjunctive radiation and chemotherapy are typically warranted.

Keywords: Burkitt, Chemotherapy, Laminectomy, Lymphoma, Spine

INTRODUCTION

Burkitt's lymphoma is a non-Hodgkin B-cell lymphoma, occurring mostly in Equatorial Africa. According to the WHO, classification is three different variants: immunodeficient-associated, endemic, and sporadic. These tumors typically impact African children, and/or those with immunodeficiency associated with HIV+. Primary "sporadic" epidural spinal lesions are rare and are usually found once the disease has already advanced (i.e., a high incidence of early metastases).[1,2] Here, we present a 63-year-old female primary "sporadic" epidural Burkitt's lymphoma who was successfully treated with tumor resection, radiation, and chemotherapy.

CASE DESCRIPTION

A 63-year-old female presented with 2-months of the low back pain (LBP) and the left lower extremity sciatica. The thoracolumbar spine MRI showed an irregular L5 osteolytic epidural lesion that was hypointense on T1-weighted studies, hyperintense on the STIR sequences, and inhomogeneously enhanced with contrast [Figures 1a-c]. Hypointense lesions also involved the L2, L3, and L4 vertebrae. The patient underwent a L4-L5 laminectomy for tumor removal (piecemeal resection), a L4-S1 transpedicular screw/rod fusion plus L2-L3 radiofrequency ablation.

Histology

The histological examination confirmed primary spinal Burkitt's lymphoma (i.e. CD20, BCL6, and PAX5 positivity, negativity for CD10, BCL2, CD23, cyclin D1, PANCK, and CK20) and a high proliferation rate (many mitotic figures and lymphoid cells with a typical starry-sky pattern).

Postoperative whole-body CT

Postoperatively, the patient's whole-body CT scan showed pleural and splenic metastatic lesions plus pathologically enlarged right hilar lymph nodes.

Postoperative course

Within 1 month postoperatively, the patient's sciatica fully recovered. She subsequently had radiotherapy, chemoradiotherapy, and 6-months later, showed >50% disease remission (i.e. on whole body CT studies).

DISCUSSION

Burkitt's lymphoma is an aggressive B-cell tumor, typically involving bone marrow and leptomeninges. Rarely patients present with primary spinal involvement.[3,4] A literature review identified 12 patients with primary epidural Burkitt's lymphoma, averaging 27 ± 20.85 years of age (range 5–69 years) [Table 1]. Patients typically present with back

Table 1: Patients' demographics.

Authors and year	Number of patients	Age (years)	Sex	Neurological symptoms	Surgical treatment	Tumor location	Chemotherapy/ radiotherapy	Outcome/ Follow-up
Sariban *et al*., 1983[8]	1	24	M	Paraplegia	Not specified	Not specified	CHOP-MTX+ RT	CNS relapse, progressive disease
Mizugami *et al*., 1987[7]	3	6, 5, 7	M, M, F	Paraparesis Paraparesis and bladder/ bowel incontinence Sensory and motor disfunction with autonomic dysfunction	Surgical decompression	T7-T11 T12-L4 T9-L2	Yes, agent not specified+ RT	Died at 20 months Died at 7 months Died at 3 months
Haddy *et al*., 1991[3]	3	34, 24, 19	M, M, F	Paraplegia	Not specified	Not specified	cyclophosphamide + vincristine+ RT	Not specified
Wilkening *et al*., 2001[10]	1	43	F	Radicular pain (S1)	Decompression and complete surgical excision	L2-L3	MTX intrathecal+ cyclophosphamide, vincristine, methotrexate, ifosfamide, adriamycin, and dexamethasone+ RT	Complete remission at two years
Daley *et al*., 2003[1]	1	13	F	Low back pain, paraparesis	Laminotomy and complete surgical excision	L1-L2	Cyclophosphamide, methotrexate, cytarabine. doxorubicin, vincristine prednisone. Intrathecal: methotrexate and cytarabine hydrocortisone	Complete remission at 68 months

(Contd...)

Table 1: Patients' demographics.

Authors and year	Number of patients	Age (years)	Sex	Neurological symptoms	Surgical treatment	Tumor location	Chemotherapy/ radiotherapy	Outcome/ Follow-up
Malani *et al*., 2006[6]	1	53	M	Low back pain, numbness, and weakness of the right leg	T3-T10 laminectomy and complete surgical excision	T4-T10	CODOX-M (cyclophosphamide/ cytarabine/doxorubicin/ leucovorin/ methotrexate/ vincristine) IVAC (cytarabine/ etoposide/ifosfamide/ methotrexate) + RT	Complete remission at 5 years
Kim *et al*., 2015[4]	1	69	F	Low back pain	L2 laminectomy and complete surgical excision	L2-L3	None	Not specified
Goitom Sereke *et al*., 2020[2]	1	8	M	Neck pain, tetraparesis, bladder and bowel incontinence	Laminectomy and decompression and complete surgical excision	C3-T2	CHOP+ intrathecal cytarabine, methotrexate, and hydrocortisone+ RT	Not specified
Costanzo *et al* 2021	1	63	F	Low back pain and left sciatica	L4-L5 laminectomy, L4-S1 osteosynthesis with transpedicular screws and rods and L2-L3 radiofrequency ablation with OsteoCool system	L4-L5	Dose adjusted EPOCH-R (Etoposide, prednisone, vincristine, cyclophosphamide, doxorubicin, and rituximab) + intrathecal MTX	>50% disease remission at 6-month follow-up

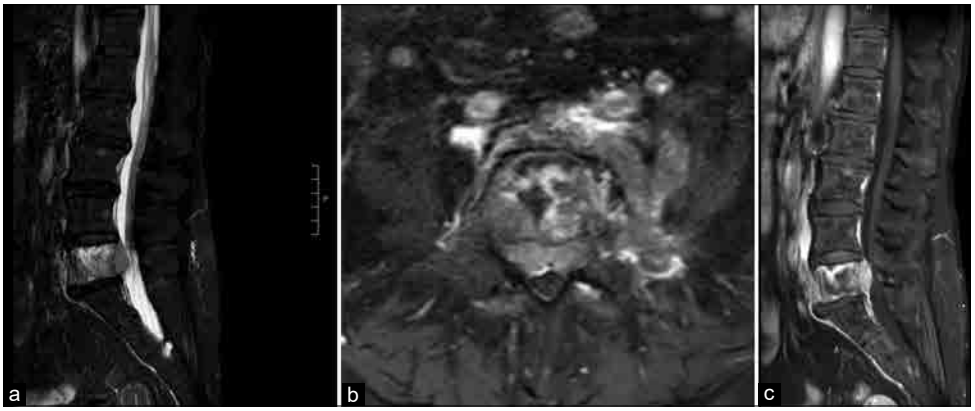

Figure 1: Preoperative thoracolumbar spine MRI showed a L5 osteolytic and epidural lesion hyperintense in sagittal STIR sequences (a) with a high and inhomogeneous post-contrast enhancement in T1-Gd axial and sagittal sequences (b and c).

pain, paresthesias, weakness, or paraparesis. The diagnostic study of choice is the enhanced MR scan Burkitt's lymphomas appear hyperintense on these contrast studies and may be single or multiple. In this case, the L5 tumor was irregular, hypointense on T1 hyperintense on STIR studies, with inhomogeneous contrast enhancement.[2,5,9,10]

Surgery

The best surgical approach to these lesions includes decompressive laminectomy with complete tumor excision, optimally followed by the adjuvant chemo-radiotherapy.[6-8] In the present case, after surgery the patient did receive chemo-

radiotherapy (i.e. Dose adjusted-EPOCH plus rituximab and intrathecal methotrexate) [Table 1].

CONCLUSION

"Sporadic" primary Spinal Burkitt's lymphoma is rare. In patients with an unknown history of lymphoma and where the spine is the primary site, surgical resection is the treatment of choice followed by radiation and chemotherapy.

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Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

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