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Primary lymphoma of bone: A case report of rare back pain presentation

Abstract—Primary lymphoma of the sacral bone is a rare myeloproliferative disease, occurring primarily after the 4th decade of life and mainly in male patients. We reported a case of sacral bone lymphoma in a 24-year-old man with low back pain and radiculopathy. Imaging showed a sacral bone lesion which later proved to be non-Hodgkin's lymphoma on the histopathologic study. Lymphoma of the sacral bone is chemoresponsive and carries a good prognosis. Sacral bone lymphoma should be considered a differential diagnosis in patients with low back pain who do not respond to analgesic treatment.

Keywords—Non-Hodgkin's lymphoma, primary bone tumour, sacral bone, chemotherapy, MRI.

1 INTRODUCTION

Lymphoma is a group of malignancies involving B cells or T cells. It usually originates from the lymph nodes. However, they may originate from any organs in the body [1]. Primary lymphoma of the sacral bone is a rare presentation [2] and usually of non-Hodgkin's lymphoma of B cell origin, presenting itself in an older age group of 50 to 60 years of age. On presentation, the commonest symptoms are low back pain with or without radiculopathy. On imaging, the tumour can mimic other tumoural lesions. However, with treatment, the prognosis of sacral lymphoma is good [3], with a five-year survival rate of 80%. We present a rare case of sacral lymphoma in our institution.

2 CASE REPORT

A 24-year-old gentleman was admitted with a history of right upper thigh and buttock pain for four months. The pain increased in severity two months before admission and radiating to both right and left lower limbs. There was intermittent numbness of both lower limbs with difficulty in ambulating at home. There was a history of loss of

appetite and loss of weight with intermittent fever. However, the patient denied any urinary or bowel incontinence. White blood cell count on admission was $26.5 \times 10^9/L$ with a platelet count of $464 \times 10^9/L$.

A plain radiograph of the pelvis was performed and showed the presence of an aggressive lytic lesion of the sacral bone (Figure 1). MRI was performed subsequently, and the examination showed a large soft tissue mass arising from the sacral bone, and it was hypointense on T1, hypo- to isointense on T2 and enhanced post-contrast (Figure 2). Core biopsy of the mass later revealed fragmented anaplastic malignant lymphoid cells arranged mainly in a diffuse pattern, in keeping with lymphoma.

The patient later underwent a course of chemotherapy, and a repeat MRI was done five months after treatment to reassess the effect of chemotherapy and the size of the tumour (Figure 3). The repeat MRI of the lumbosacral spine showed a reduction in size of the tumour indicating a good response to chemotherapy with resolving symptoms.



Figure 1: Plain radiograph of the pelvis on admission show ill-defined lytic lesion with wide zone of transition involving the sacral bone. These findings are suggestive of an aggressive bone lesion involving the sacrum.

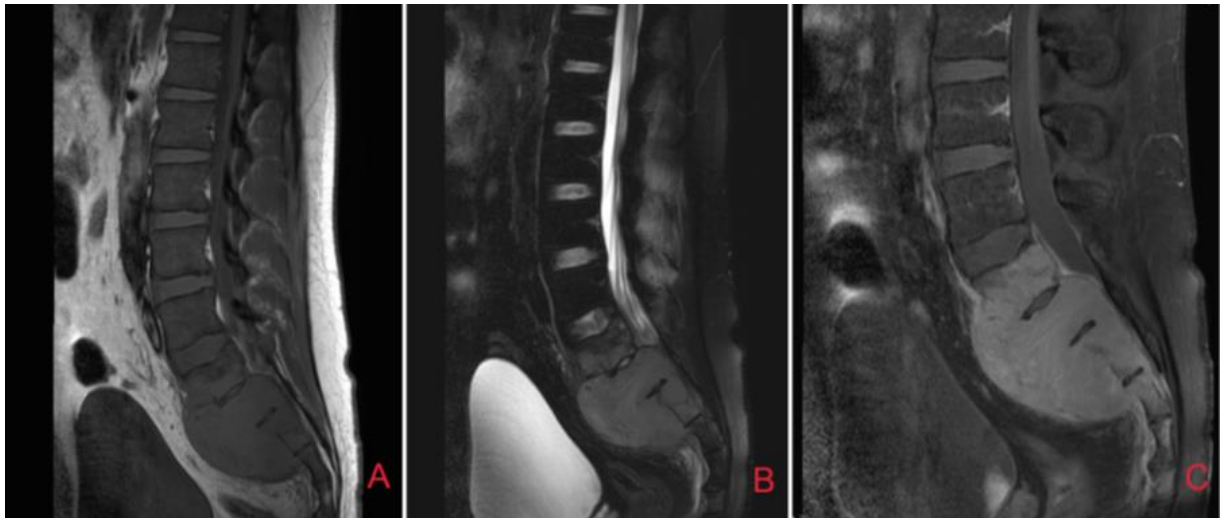


Figure 2: MRI of the lumbosacral spine showing a lesion which is hypointense on T1 [A], hypo- to isointense on T2 [B] and enhanced post contrast [C].

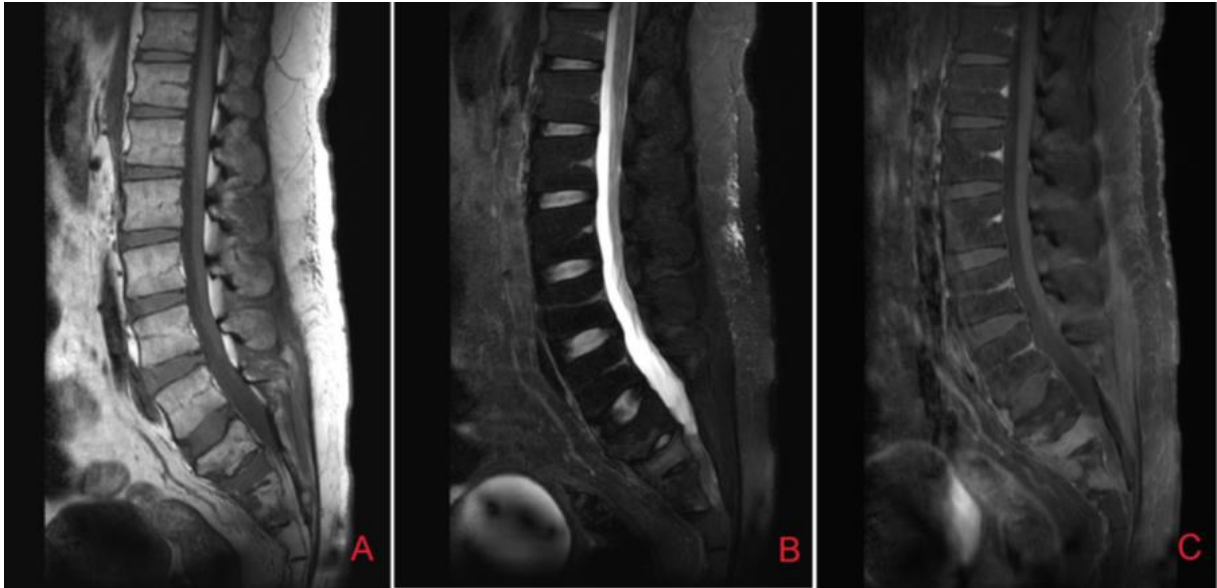


Figure 3: Repeat MRI of the lumbosacral spine in T1-weighted [A], T2-weighted [B] and post contrast [C] 5 months post chemotherapy shows reduction in size to the tumour indicating good response to treatment.

3 DISCUSSION

Low back pain is one of the common complaints encountered in primary care setting [4]. Most cases are self-limited with no causes identified and attributed to muscular or ligamentous injury [5]. It can be accompanied by sciatica, in which more extensive workup should be performed and usually caused by herniated intervertebral disk, spinal stenosis, and cauda equina syndrome. Apart from degenerative disc disease of the spine, trauma, infections and neoplasm can cause low back pain [6]. The commonest cause of sacral neoplasm is metastases, while the commonest cause of primary neoplasm of the sacrum is chordoma [3].

Lymphoma with secondary involvement of the axial skeleton is much more common than primary skeletal lymphoma [7]. Primary bone lymphoma is rare, occurring in 3% of all primary malignant bone tumours and 5% extranodal lymphoma [8]. The disease can occur at any age with an age-onset of more than 10 years old and commonest in the age group of over 40 to 60 years old and more common in male patients [9]. Commonly the disease affects the femur and pelvic bones and rarely involve the spinal epidural [10].

Our patient falls into an earlier age group than commonly described in patients with primary

sacral lymphoma. Although some literature described the age of onset could be as early as 20 years, the majority presents in a much older age group [11].

The most typical presentation of patients with primary lymphoma of the sacrum is low back pain with radiating pain to the lower limbs. However, patients can also be asymptomatic at presentation [12]. The symptoms worsen as the spinal cord is compressed, followed by extremity weakness, paresis and paralysis.

Plain radiographs usually reveal ill-defined lytic lesion involving the sacrum with a wide zone of transition. Some literature describes the lesion as mixed moth-eaten destruction with patchy areas of sclerosis, which is non-specific [7]. MRI examination shows hypointense signal intensity on T1 weighted images and T2 weighted images, which are likely due to the presence of fibrosis in patients with non-Hodgkin's lymphoma [13]. As in Hodgkin's lymphoma, more inflammatory cells and fluid show hyperintense signal on T2 weighted images [14].

In most cases, sacral lymphomas are chemoresponsive to treatment and parallel with the systemic response to chemotherapy. Evaluation of response is done with MRI. MRI also enables to show the complication that arises as well as the involvement of the spinal cord.

Generally, patients with primary bone lymphoma treated with a combination of radiation therapy and chemotherapy do well. A total 5-year survival rate of 83% was seen in a study by Tondini et al. [15]. The overall 5-year survival rate is better than that for most other primary osseous malignancies. For this reason, recognition of lymphoma as a possible cause for the described radiologic findings is essential [16].

4 CONCLUSION

Primary sacral lymphoma should be considered a differential diagnosis in a patient with low back pain that is not responding to analgesics. Bone tumour should be considered in patients with these kinds of presentations. Although rare, early diagnosis would provide a better prognosis for the patient.

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CONSENT FOR PUBLICATION

Written consent was granted from the patient for the publication of this case report and the accompanying images. Institutional Review Board approval is not required at the authors' institutions for the presentation of a single case report.

REFERENCES

- [1] Eeles RA, O'Brien P, Horwich A, Brada M. Non-Hodgkin's lymphoma presenting with extradural spinal cord compression: functional outcome and survival. *Br J Cancer* [Internet]. 1991;63[1]:126–9. Available from: <http://www.ncbi.nlm.nih.gov/pubmed/1989650> %5Cnhtp://www.pubmedcentral.nih.gov/articlerender.fcgi?artid=PMC1971629
- [2] Epelbaum R, Haim N, Ben-Shahar M, Ben-Arie Y, Feinsod M, Cohen Y. Non-Hodgkin's lymphoma presenting with spinal epidural involvement. *Cancer*. 1986;58[9]:2120–4.
- [3] Liu JK, Kan P, Schmidt MH. Diffuse large B-cell lymphoma presenting as a sacral tumor. Report of two cases. *Neurosurg Focus* [Internet]. 2003;15[2]:E10. Available from: <http://www.ncbi.nlm.nih.gov/pubmed/15350041>
- [4] Lateef H, Patel D. What is the role of imaging in acute low back pain? *Curr Rev Musculoskelet Med*. 2009;2[2]:69–73
- [5] Last AR, Hulbert K. Chronic low back pain: Evaluation and management. *Am Fam Physician*. 2009;79[12]:1067–74
- [6] Roug IK, McCartney LB. Metastatic non-Hodgkin lymphoma presenting as low back pain and radiculopathy: A case report. *J Chiropr Med* [Internet]. 2012;11[3]:202–6. Available from: <http://dx.doi.org/10.1016/j.jcm.2012.05.008>
- [7] Chotai N, Dutta R. Primary intradural Hodgkin's lymphoma in lumbosacral spine: A rare location. *Clin Neuroradiol*. 2010;20[4]:247–9
- [8] Vincent J.M., Ng Y.Y., Norton A.J. AP. Case Report : Primary Lymphoma of Bone- MRI Appearances With Pathological Correlation. *Clin Radiol* [Internet]. 1992;45[6]:407–9. Available from: [http://www.clinicalradiologyonline.net/article/S0009-9260\(05\)81002-5/pdf](http://www.clinicalradiologyonline.net/article/S0009-9260(05)81002-5/pdf).
- [9] Schiller AL, Wang CC, Suit HD. Primary Lymphoma of Bone The Relationship of Morphologic Diversity to Clinical Behavior. *Cancer* [Internet]. 1982;50[5]:1009–1014. Available from: [http://onlinelibrary.wiley.com/doi/10.1002/1097-0142\(19820901\)50:5%3C1009::AID-CNCR2820500532%3E3.0.CO;2-0/abstract](http://onlinelibrary.wiley.com/doi/10.1002/1097-0142(19820901)50:5%3C1009::AID-CNCR2820500532%3E3.0.CO;2-0/abstract)
- [10] Theodorou DJ, Theodorou SJ, Sartoris DJ, Haghighi P, Resnick D. Delayed diagnosis of primary non-Hodgkin's lymphoma of the sacrum. *Clin Imaging* [Internet]. 2000;24[3]:169–73. Available from: <http://www.ncbi.nlm.nih.gov/pubmed/11150687>
- [11] Resnick D. *Diagnosis of Bone and Joint Disorders*. 3rd ed. 3rd ed. Philadelphia, Pa: W.B. Saunders & Co.; 1995. 4496 p.
- [12] Wong ET, Portlock CS, O'Brien JP, DeAngelis LM. Chemosensitive epidural spinal cord disease in non-Hodgkins lymphoma. *Neurology* [Internet]. 1996;46[6]:1543–7. Available from: <http://ovidsp.ovid.com/ovidweb.cgi?T=JS&PAGE=reference&D=med4&NEWS=N&AN=8649545>
- [13] Stiglbauer R, Augustin I, Kramer J, Schurawitzki H, Imhof H, Radaszkiewicz T. MRI in the diagnosis of primary lymphoma of bone: correlation with histopathology. *J Comput Assist Tomogr*. 1992;16[2]:248–53.
- [14] Hermann, G., Klein, M. J., Abdelwahab, I. F. & Kenan S. MRI appearance of primary non-Hodgkin's lymphoma of bone. *Skeletal Radiol* [Internet]. 1997;26[11]:629–32. Available from: <https://www.ncbi.nlm.nih.gov/pubmed/9428068#>
- [15] Tondini C, Zanini M, Lombardi F, Bengala C, Rocca A, Giardini R, et al. Combined modality treatment with primary CHOP chemotherapy followed by locoregional irradiation in stage I or II histologically aggressive non-Hodgkin's lymphomas. *J Clin Oncol Off J Am Soc Clin Oncol*. 1993 Apr;11[4]:720–5.
- [16] Krishnan A, Shirkhoda A, Tehranzadeh J, Armin AR, Irwin R, Les K. Primary bone lymphoma: radiographic-MR imaging correlation. *Radiographics* [Internet]. 2003;23[6]:1371–7. Available from: <papers3://publication/doi/10.1148/rg.236025056>