IJHOSCR

Case Report

International Journal of Hematology-Oncology and Stem Cell Research

Infected Primary Bone Mantle Cell Lymphoma with Multiple Vertebral Involvement

Rebeh Bougossa[1], Fatma Larbi[1], Asma Ben Mabrouk[1], Olfa Jomaa[2], Jihene Chelli[1]

[1]Department of Endocrinology and Internal Medicine, Tahar Sfar University Hospital, Mahdia, Tunisia
[2]Department of Rheumatology, Tahar Sfar University Hospital, Mahdia, Tunisia

Corresponding Author: Rebeh Bougossa, Department of Endocrinology and Internal Medicine, Tahar Sfar University Hospital, Mahdia, Tunisia
Tel: +21621923696
E-mail: rebeh.gos@gmail.com

Received: 19, Feb, 2024
Accepted: 03, Aug, 2024

ABSTRACT

Primary vertebral lymphoma is an exceedingly rare entity. We hereby report a case of a 67-year-old male who presented to our department with fever, weight loss, and progressively worsening lower back pain radiating to the right hip. Physical examination showed pain on percussion of the dorsal and lumbar spine and tenderness on palpation of the right upper thigh area. The radiographic findings revealed numerous vertebral lesions with a right psoas abscess extending to the right upper thigh. Intravenous antibiotic therapy was initiated and the patient underwent an incision and drainage of the psoas abscess with a favourable outcome. However, given the suspicious imaging findings of the osseous lesions suggestive of malignancy, a vertebral biopsy was performed and yielded histo-pathological findings consistent with bone mantle cell lymphoma. To the best of our knowledge, this is the first case of an infected primary bone mantle cell lymphoma with multiple vertebral involvement. The diagnosis is challenging and can be confused with other diseases.

Keywords: Mantle cell lymphoma; Vertebrae; Infected tumor

INTRODUCTION

Lymphomas are a group of malignancies of B, T or natural killer cells that most commonly originate from lymph nodes, but any other primary extranodal sites may be involved. The primary bone lymphoma (PBL) is an exceedingly rare entity, accounting for approximately 3% to 7% of all primary bone tumors and less than 5% of the extranodal lymphomas[1]. The prevalence of primary vertebral sites is estimated at only 1.7% amongst all PBLs[2]. The clinical and radiological features of PBLs are usually unspecific and the diseases can be misdiagnosed as other bone tumors or chronic osteomyelitis. The diagnosis of PBL is histologic. In order to provide insight about this rare malignancy, we report, in this paper, an unusual case of an infected primary vertebral lymphoma in a 67- year-old man.

To the best of our knowledge, this is the first case of an infected primary bone mantle cell lymphoma with multiple vertebral involvement.

Case presentation

A 67 -year-old male presented to the Department of Infectious Diseases at Tahar Sfar University Hospital, with a two- month history of fever and weight loss, and a progressively worsening lower back pain radiating to the right hip without muscular weakness or paresthesia over the last 3 weeks.

His past medical history included arterial hypertension and dyslipidemia. He was non diabetic and he did not smoke or consume any alcohol. He reported the use of paracetamol and non-steroidal anti-inflammatory drugs, but without any relief.

On physical examination, the patient was febrile (T°=39°C), the respiratory and cardiovascular

examinations were unremarkable. Pain on percussion of the dorsal and lumbar spine, and a tenderness on palpation of the right upper thigh area were noted. The muscular force was normal for upper and lower limbs, but the range of movement of his right hip was limited due to pain. Patellar reflex and Achilles tendon reflex were normal and Babinski's sign was negative on both sides. No superficial lymphadenopathy or hepatosplenomegaly were palpable.

Serum tests revealed the presence of leukocytosis (17000/mm³) and high level of C reactive protein (150mg/L). Haemoglobin and platelet count were within normal limits. Renal and hepatic functions, serum electrophoretic study and tumor markers were normal. Radiographs of chest and dorso-lumbar spine were normal. Computed Tomography (CT) and magnetic resonance imaging (MRI) of the spine showed an infiltration of the whole spine with numerous osseous lesions that were hypointense on T1 and slightly hyperintense on T2 weighted images with no evidence of fracture or cord compression. There was also, a 30 ×4 cm right psoas abscess extending to the right upper thigh (Figure 1).

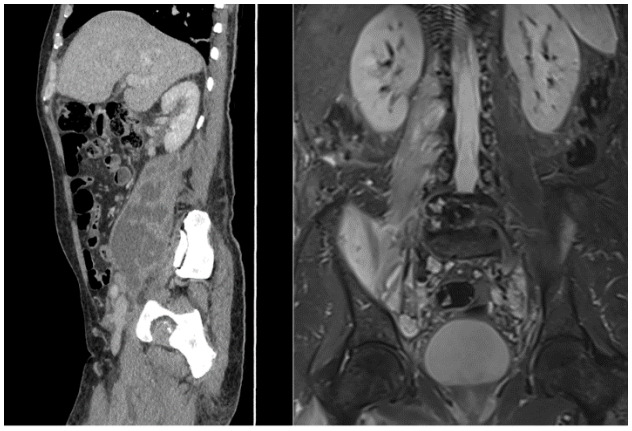

Figure 1. The right psoas abscess extending to the right upper thigh on the imaging exams

Empirical intravenous cefazolin was initiated, and after 3 days, the patient underwent an incision and drainage of the psoas abscess under general anaesthesia. The bacterial cultures taken during the operation did not show the growth of any microorganism. Two weeks later, all laboratory results were within normal limits and a repeat CT demonstrated a partial resolution of the right psoas abscess.

The initial antibiotic regimen was switched to four weeks of ceftazidime and teicoplanin with good clinical and radiological outcomes. However, in view of the suspicious imaging findings of the osseous lesions suggestive of malignancy, a vertebral biopsy was performed under CT scan guidance and yielded histo-pathological findings consistent with bone mantle cell lymphoma. Bacterial and fungal cultures of the biopsy specimen did not show any microorganism growth and the acid-fast stain and tuberculosis polymerase chain reaction were negative. CT scans of chest, abdomen and pelvis did not show any mass or nodule or significant lymphadenopathy suggesting the dissemination of the disease. Thus, the patient was diagnosed with primary vertebral mantle cell lymphoma with superinfected bacterial osteomyelitis and he was referred to the department of clinical haematology.

DISCUSSION

PBL was first described by Oberling in 1928 [3] and then, in 1939 the disease was recognized by Parker and Jackson as a distinct entity; a primary reticulum cell sarcoma of the bone [4].

In the literature, the exact definition of PBL has been controversial over time. According to the 2013World Health Organization classification of soft tissues and bone tumors , PBL was defined as either a single osseous lymphoma with or without regional lymph node involvement, or multiple osseous lesions without nodal or extranodal disease, also called "multifocal osseous lymphoma" or "polyostotic lymphoma"[5]. However, in advanced stages of the disease, it may be difficult to distinguish between PBL and secondary bone lymphoma.

The causes of PBL are still not well-Known. Some studies have identified correlations between PBL and other bone diseases (e.g. Paget's disease), viral infections (e.g. Human immunodeficiency virus infection), autoimmune disorders and organ transplantation. However, there are no well-established risk factors for the development of this malignancy [6].

Our patient was a 67 -year-old male. He had no history of autoimmune disorders or bone diseases. The majority of patients were diagnosed with PBL between the 5th and 6th decades with a slight male predominance[7]. There is no much data to conclude to any geographical or racial disparities in this malignancy. Any part of the skeleton can be involved in PBL, but the axial skeleton is more commonly affected than the appendicular skeleton [8]. The main clinical symptoms of PBL are local pain and swelling due to the extension of the disease to the soft tissues. Neurological symptoms, such as spinal cord compression and pathological fractures, can be noted. However, systemic "B-type" symptoms such as fever, night sweats and weight loss are relatively uncommon[7, 8]. As reported by retrospective study including 109 PBL , the median duration of symptoms was 8 months, but it could exceed one year in some cases [7]. In our case, the symptoms were more likely related to the vertebral osteomyelitis than PBL, because there was a clinical resolution after the treatment of the pyogenic osteomyelitis and the psoas abscess. Here, the superinfection can be explained by the haematological spread of microorganisms to areas of tumor necrosis. The bacterial cultures did not show any microorganism growth because they were probably decapitated by antibiotic therapy.

The radiographic findings are unspecific in PBL. On bone X-ray, lesions are usually lytic, but osteoblastic lesions can be noted and mixed patterns can even coexist in the same bone. CT is used to delineate cortical destructions and extraosseous lesions. However, MRI provides clearer details on the invasion and the extent of the disease than CT especially when the central nervous system is involved. The signal intensities in the lesions are often hypointense or isointense on T1 weighted and hyperintense on T2 weighted MRI[9]. The clinical and radiological findings are unspecific in PBL and the diagnosis can be confused with spinal infection or tuberculosis especially when the lesions are localized in the lateral vertebrae or spinal epidural. Moreover, PBL can be misdiagnosed with other bone tumors. In our case, in addition to the diagnosis of pyogenic vertebral osteomyelitis complicated by a psoas abscess, a malignant infiltration of the spinal bone was suspected in the imaging findings, therefore a vertebral biopsy was carried out and the histopathological findings confirmed the additional diagnosis of primary vertebral mantle cell lymphoma. The histopathological findings of the bone or the adjacent lymph node biopsy form the foundation of the diagnosis of PBL. Diffuse large B-cell lymphoma is the most common histologic subtype of primary and secondary lymphoma infiltrating the bone accounting for to 80% of all PBL[8]. While the rest of the subtypes such as mantle cell, follicular, marginal zone, lymphoplasmocytic, anaplastic large cell, Natural killer/T-cell, Burkitt and Hodgkin lymphomas [8]. It is recommended to reduce the amount of resected tissue in excisional biopsies in order to avoid the risk of pathological fractures.

To the best of our Knowledge, this is the first case of a multifocal primary vertebral mantle cell lymphoma with superimposed pyogenic osteomyelitis complicated with a psoas abscess.

There is no standard therapy or guideline for PBL, as all previous studies have been retrospective and most of them have been case reports. Therapy in general is multimodal and includes surgery, radiotherapy, chemotherapy and rituximab[10].

CONCLUSION

This case highlights a unique presentation of an infected primary bone mantle cell lymphoma with multiple vertebral involvement. As no clinical or radiological findings are specific to PBL, the diagnosis is challenging and can be confused with other diseases. Therefore, physicians should be aware of this possible diagnosis, especially in front of unusual clinical or radiological features of vertebral osteomyelitis, and then the diagnosis of PBL is histologic.

Role of the funding source

This research did not receive any specific grants from funding agencies in the public, commercial, or not-for-profit sectors.

Informed consent

The informed consent was obtained from the patient for publication of the details and accompanying images.

REFERENCES

1. Freeman C, Berg JW, Cutler SJ. Occurrence and prognosis of extranodal lymphomas. Cancer. 1972; 29(1):252-60.
2. Rathmell AJ, Gospodarowicz MK, Sutcliffe SB, et al. Localized extradural lymphoma: survival, relapse pattern and functional outcome. The Princess Margaret Hospital Lymphoma Group. Radiother Oncol. 1992; 24(1):14-20.
3. Oberling C. Les réticulosarcomes et les reticuloendothéliosarcomes de la moelle osseuse (sarcomes d'Ewing). Bull Assoc Fr. Etude Cancer. 1928; 17: 259-296.
4. Parker F Jr, Jackson H Jr. Primary reticulum cell sarcoma of bone. Surg Gynecol Obstet.1939; 68:45-53.
5. Rosenberg AE. WHO Classification of Soft Tissue and Bone, fourth edition: summary and commentary. Curr Opin Oncol. 2013; 25(5):571-3.
6. Bindal P, Desai A, Delasos L, et al. Primary Bone Lymphoma: A Case Series and Review of Literature. Case Rep Hematol. 2020;2020:4254803.
7. Müller A, Dreyling M, Roeder F, et al. Primary bone lymphoma: Clinical presentation and therapeutic considerations. J Bone Oncol. 2020;25:100326.
8. Messina C, Christie D, Zucca E, et al. Primary and secondary bone lymphomas. Cancer Treat Rev. 2015; 41(3):235-46.
9. Shen G, Su M, Liu B, et al. PET/CT Imaging for Solitary Primary Bone Lymphoma of Thoracic Vertebra. Clin Nucl Med. 2018; 43(11):857-859.
10. Zhou HY, Gao F, Bu B, et al. Primary bone lymphoma: A case report and review of the literature. Oncol Lett. 2014;8(4):1551-1556.