PAGET’S DISEASE OF BONE DIAGNOSED ON SPECT/CT: A CASE REPORT

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Abstract

I reported a case of a seventy-five-year-old woman with a backache and pain in left femur. Magnetic resonance imaging (MRI) of back, pelvis and hips showed bone marrow lesion suggesting bone metastasis. The patient was admitted to nuclear medicine department of Krasnoyarsk Regional Clinical Oncology Center. Single-photon emission computed tomography combined with computed tomography (SPECT/CT) of the skeletal system together with several laboratory tests (alkaline phosphatase, calcium, phosphorus), provided grounds for the diagnosis of Paget’s disease. The patient was qualified for treatment to the Rheumatologist.

Keywords

Paget’s disease of bone • bone pain • metabolic bone diseases • SPECT/CT • alkaline phosphatase

Introduction

Paget’s disease of bone (PDB) is a chronic progressive disease of the bone of uncertain etiology, characterized initially by an increase in bone resorption, followed by a disorganized and excessive formation of bone, leading to pain, fractures, and deformities. PDB is the most common metabolic bone diseases after osteoporosis [1]. The disease predominately affects elderly and male patients [2]. The article describes the utility of single photon emission computed tomography combined with computed tomography (SPECT/CT) to rule out metastatic bone disease in a patient Paget’s disease mimicking multiple skeletal metastases.

Case report

A 75-year-old woman, presented with a history of backache and pain in left femur. She also complained of excessive worries, tension, and having sleep disturbance. There was no past history of diabetes, hypertension, tuberculosis, polyarthritis, or any other significant illness. The family history was not contributory. Magnetic resonance imaging (MRI) of back, pelvis and hips showed bone marrow lesion suggesting skeletal metastases. The patient was admitted to the nuclear medicine department of the Krasnoyarsk Regional Clinical Oncology Center. Whole-body bone scintigraphy was performed two hours after intravenous administration of technetium-99m methylene diphosphonate (MDP). Images revealed multiple skeletal lesions (“hot spots”) involving skull, multiple vertebrae, pelvis on both sides, and upper half of the right femur (Figure 1).

Figure 1. Planar bone scintigraphic images showing increased tracer uptake in skull, multiple vertebrae, pelvis on both sides and upper half of the right femur.
Biochemical tests revealed normal serum calcium: 2.3 mmol/L (2.2-2.55 mmol/L) and serum phosphorus: 1.23 mmol/L (0.81-1.45 mmol/L), but markedly raised serum alkaline phosphatase (SAP) – 683 IU/L (35-105 IU/L). Correlating the clinical, radiologic and biochemical findings a final diagnosis of PDB was established. Patient was qualified to the Rheumatologist for treatment.

Discussion

PDB is a chronic, non-inflammatory, localized bone-remodeling disorder that affects widespread, non-contiguous areas of the skeleton. PDB is the second most common bone disease after osteoporosis [3]. This disease is relatively common in older people, occurs in approximately 3-4% of the population aged over 50 years with a slight male gender predilection. PDB can be monostatic or polyostotic in nature depending on the number of bones involved. Most commonly involved bones are the pelvic girdle, spine, lumbar region, thoracic region as well as and cervical and skull bones. Most cases are asymptomatic in nature but symptomatic cases may lead to various manifestations such as arthritis, bone pain, pathological fractures, bowing of legs and kyphosis [4].

PDB is diagnosed primarily by radiological examinations. Early in the course of the disease, lytic activity predominates, causing focal osteolytic lesions. Subsequently, areas of sclerosis develop, leading to the characteristic appearances of mixed...
lytic and sclerotic areas, thickened trabecula, bone expansion, cortical thickening, and deformity. A radioisotope bone scan may be recommended in all patients as part of the initial diagnostic assessment to determine the distribution of the disease [5]. Different biochemical markers of bone remodeling that are increased in PDB play a useful role in the diagnosis of the disease. The markers of bone resorption, which are increased, are: urinary hydroxyproline, serum N-telopeptide of type I collagen, serum C-telopeptide of type I collagen and serum deoxypyridinoline cross-links of type I collagen. Markers of bone formation that are elevated are: SAP, serum bone-specific alkaline phosphatase, osteocalcin, serum N-terminal propeptide of type I collagen [6]. There are several treatment regimens for patients with PDB. Bisphosphonates have been proven as the first-line treatment option, secondary to its influence in bone remodeling. The most commonly used bisphosphonates in the management of Paget’s disease are pamidronate, etidronate, zolendronic acid, alendronate and riserdronate [7]. Calcitonin is usually a second-line treatment. This drug is assisted in bone absorption. Supplements such as calcium and vitamin D have been known to provide some symptomatic benefit. Pain management achieved with either nonsteroidal anti-inflammatory drugs (NSAIDs) or acetaminophen [8]. The general outlook for patients with PDB is good, especially if treatment started before major changes have occurred in the bones. There is no cure for PDB, but the disorder can be controlled from progressing. Patients with severe polyostotic Paget disease have a less favorable prognosis than those with monostotic disease [9]. Only one study, however, has specifically addressed mortality associated with PDB. In that study, the British General Practice Research Database identified 2465 patients diagnosed with PDB from 1988 to 1999: retrospective review indicated that 5-year survival was 67% in patients with Paget’s disease compared with 72% in control patients [10]. The prognosis is extremely unfavorable if the patient has any type of sarcomatous degeneration, especially if there is multicentricity. The 5-year survival rate of Paget’s sarcoma is approximately 10%, much worse than that of conventional osteosarcoma, which has increased to nearly 70% with the improvement of neoadjuvant chemotherapy. Most tumors show a poor response to standard chemotherapy regimens used for conventional osteosarcoma [11, 12].

**Conclusion**

This case presented utility of SPECT/CT uniting isotope bone scan and CT-scan in diagnostics of PDB which can be confirmed by elevated alkaline phosphatase levels in serum which was positive in our patient, too.

**Conflict of Interest Statement**

Author declares no conflict of interests for this article

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