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Case Report

^{99m}Tc-HMDP Bone Scintigraphy and Paget's Disease: About 7 Cases in the Nuclear Medicine Department of Idrissa Pouye General Hospital in Dakar

El Hadji Amadou Lamine Bathily[1, 2, *], Olatounde Herbert Fachinan[1], Ousseynou Diop[2, 3], Mamoudou Salif Djigo[1, 2], Gora Thiaw[1], Kalidou Gueye [1], Mohamed Chekhma[1], Boucar Ndong[2, 3], Omar Ndoye[2], Mamadou Mbodj[1, 2]

[1]Nuclear Medicine Department, Idrissa Pouye General Hospital (HOGIP), Dakar, Senegal

[2]Biophysical Laboratory, Faculty of Medicine, Pharmacy and Odontology (FMPO), Cheikh Anta Diop University (UCAD), Dakar, Senegal

[3]Nuclear Medicine Department, Hospital Dalal Jamm, Dakar, Senegal

Email address:

bathilyssd@yahoo.fr (El Hadji Amadou Lamine Bathily)

[*]Corresponding author

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Abstract: Paget's disease is a chronic disorder of the adult skeleton characterized by focal dys-regulation of bone remodelling. The aim of this study was to demonstrate the contribution of bone scintigraphy to the diagnosis of Paget's disease. The study was observational, cross-sectional and descriptive. It included patients who underwent bone scintigraphy in the nuclear medicine department of Dakar's Idrissa Pouye General Hospital from January 2020 to June 2023, and whose results suspected or confirmed Paget's disease of the bone. We collected a total of seven (07) patients during the study period, six (06) of whom were male with an average age of 67.42 years. Two (02) reasons justified the performance of scintigraphy in these patients. These were suspicion of Paget's disease and assessment of bone extension of prostate cancer with incidental discovery of Paget's disease. Two patients (28.57% of cases) had the monostotic form and five (71.43% of cases) had the polyostotic form. All lesions found on bone scans were hyper-fixating. Three (03) patients showed bone deformities, notably deformities of the long bones, and two (02) had typical osteocondensing (hyperfixation) images of the vertebrae, giving them a "Mickey Mouse" or clover appearance. Whole-body bone scintigraphy has made an important contribution to the diagnosis and assessment of the extent of Paget's disease, with a favourable dosimetric ratio.

Keywords: Paget's Disease, Bone Scintigraphy, Dakar

1. Introduction

Paget's disease is a progressive metabolic osteopathy characterized by focal areas of increased bone turnover that can affect any bone [1]. It generally affects the elderly, and men are more at risk than women [2, 3]. In Europe, around 2% of adults over 55 are affected, but it remains rare in Africa [4, 5].

The disease has a predilection for the pelvis, spine, skull and long bones [1, 6, 7]. Its etiology is only partially understood, and includes both genetic and environmental factors [6], as well as infectious factors, especially viral [1]. It is most often manifested by bone pain, deformities, fractures or other complications, notably neurosensory [1], but can also remain asymptomatic and be discovered fortuitously on medical imaging (radiography, isotope exploration) or biology during para-clinical examinations carried out to explore another pathology [8].

Radionuclide bone scintigraphy is considered a useful technique for diagnosing Paget's disease and assessing its

extent, due to its high sensitivity and ability to scan the entire skeleton in a single pass with a favourable dosimetric ratio [1, 5].

The aim of the study was to analyse the results of bone scintigraphy performed in the nuclear medicine department of Idrissa Pouye General Hospital (IPGH) in Dakar, as part of the diagnosis of Paget's disease.

2. Methodology

2.1. Study Method

This was an observational, cross-sectional and descriptive study. It included patients who underwent bone scintigraphy at the nuclear medicine department of Idrissa POUYE General Hospital in Dakar from January 2020 to June 2023, and whose results suspected or confirmed Paget's disease of the bone.

To collect and exploit the data, we used the patients' bone scan records from the software database (InterViewXP / Médiso), and the physical records (clinical observation sheets) of each patient included in the study.

Sampling was exhaustive. All patients who had undergone a bone scan and whose results suspected or confirmed Paget's disease were included. For each patient file, the data collected were related to the socio-clinical, biological and scintigraphic data. These data were transcribed onto a data processing form designed for the study.

2.2. Three-Phase (03) Bone Scintigraphy Technical

For whole skeleton isotope exploration, a bisphosphonate derivative, methylene-hydroxide bisphosphonate (MHBP) labelled with metastable technetium 99 (^{99m}Tc) was injected intravenously with an activity of 8 to 10 MBq/kg, not to exceed 1200 MBq.

Early dynamic acquisition (angiographic phase) was performed just after injection of the radiotracer, using a SPECT Mediso double-headed gamma camera (anterior face-posterior face) equipped with a high-resolution low-energy parallel collimator focused on the suspect and contralateral regions.

Late bone acquisition was performed in patients lying supine on the examination table, three hours after injection, using a whole-body scintigraphy by the same gamma camera at a speed of 15 cm per minute.

3. Results

3.1. Socio-Demographic Data

We enrolled a total of seven (07) patients during the study period, six (06) of whom were male. The mean age was 67.42 years, with extremes of 43 and 88 years.

3.2. Clinical-Biological Data

3.2.1. Clinical Manifestations

Two (02) cases (28.57%) were asymptomatic. Of the five (05) symptomatic cases, four (04) had osteoarticular pain, two (02) had pathological fractures, two (02) had difficulty walking and one (01) had a neurosensory complication (flaccid paralysis).

3.2.2. Biological Abnormalities

Total alkaline phosphatase was measured in three patients, with elevated levels in two (02) (above 130 IU/L).

3.3. Scintigraphy Data

3.3.1. Reason for Scintigraphy

Two (02) reasons justified the performance of scintigraphy in the patients. Three (03) patients were referred for suspected Paget's disease and four (04) patients were referred for assessment of bone extension of prostate cancer, and the discovery of Paget's disease was incidental in these cases.

3.3.2. Angiographic Phase

At the angiographic stage, hyperaemia of the affected areas compared with contralateral areas was observed in five (05) patients (71.42% of cases).

3.3.3. Disease Form

Two patients (28.57% of cases) were monostotic and five (71.43% of cases) polyostotic.

3.3.4. Lesion Site

Five (05) patients had lesions on the hipbone, three (03) on the long bones, notably the femur and humerus, and two (02) on the spine.

3.3.5. Type of Lesion

All lesions found on bone scintigraphy were hyperfixed. Three (03) patients showed bone deformities, in particular deformities of the long bones and two (02) cases of typical osteo-condensing (hyperfixation) images of the vertebrae (figure 1), giving a "Mickey Mouse" appearance.

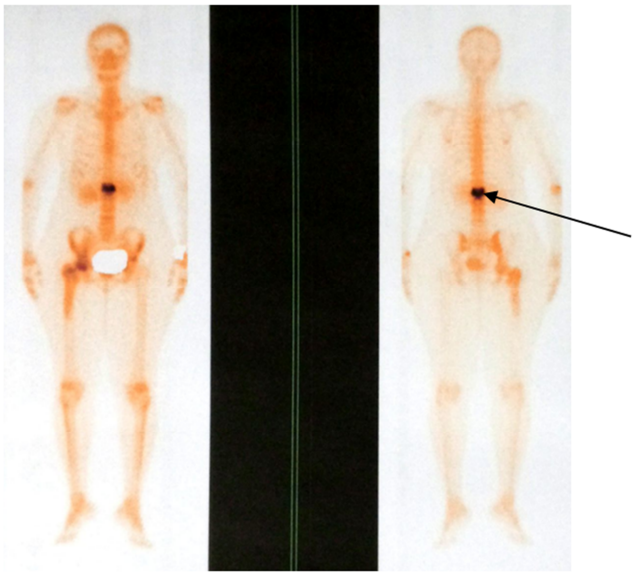

Figure 1. *Whole-body bone scintigraphy with ^{99m}Tc-MHBP, left anterior and right posterior in a 43-year-old woman.*

It shows intense, homogeneous hyperfixation of the first lumbar vertebra (Mickey Mouse appearance) and discrete hyperfixation of the right coxo-femoral joint and the upper half of the homo-lateral femur, consistent with Paget's disease.

3.3.6. Interpretation of a Case Study

This was an 88-year-old patient referred by the rheumatology department on suspicion of Paget's disease, in the context of a pathological right per-trochanteric fracture. Clinical examination revealed pain in the right hip and thigh, with difficulty in prolonged walking. Biochemistry revealed an elevated total alkaline phosphatase level of 267 IU/L (35 - 130 IU/L). Standard radiography revealed heterogeneous condensation of the right femur, with thickening of the cortices. Whole-body bone scintigraphy performed after injection of 630 MBq of ^{99m}Tc-MHBP and using a gamma camera objectified:

In the angiographic and tissue phase: hyperaemia of the right hip and upper third of the right femur, with an average vascular ratio of 1.34 compared with the contralateral side (figure 2).

Late bone stage: hyper-fixations of the right femur, right hemi-basin, the sacrum and the fifth cervical vertebra (C5) associated with curvature of the right femur (figure 3).

This scintigraphic appearance in the patient's clinical-biochemical context was compatible with polyostotic Paget's disease.

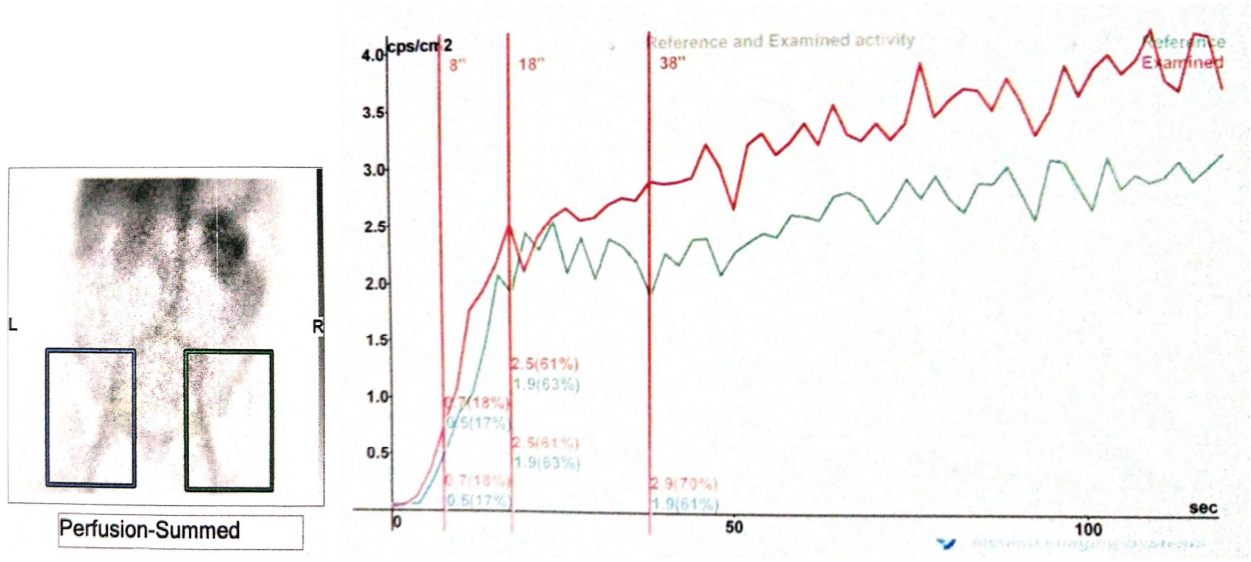

Figure 2. *Perfusion curves for 2 hips and 2 femurs, right side in red and left side in green.*

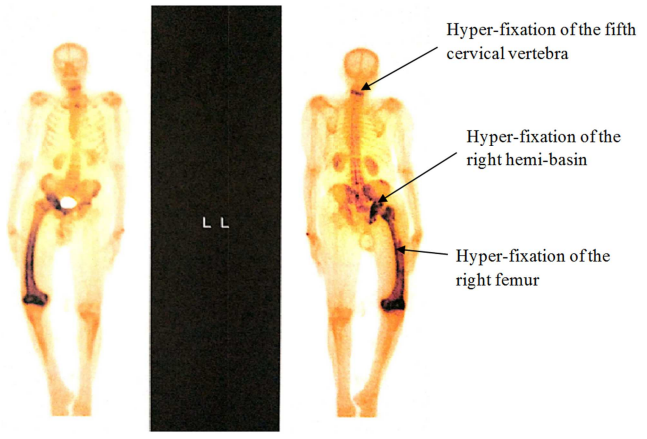

Figure 3. *Whole-body bone scintigraphy with ^{99m}Tc-MHBP, left anterior and right posterior, Paget's disease affecting several bones.*

4. Discussion

Paget's disease remains rare in sub-Saharan Africa, affecting less than 0.1% of the population [5]. The rarity of the disease in Africa was highlighted during the study period, with only 7 cases received over almost 4 years.

It occurs after the age of 55, preferably in males [2, 4, 5], which is consistent with our sample. We found an average age of 67.42 years, with extremes of 43 and 88 years, and a sex ratio of 6.

Of the five symptomatic cases, four (80%) had osteoarticular pain. The literature reports that pain frequently occurs in pelvic and lower limb involvement, and is associated with pathetic coxopathy with extensive bone remodelling [6, 8]. In contrast, two (02) cases (28.57%) were asymptomatic in our study. The discovery of Paget's disease was fortuitous in these cases. Cases of fortuitous discovery have been reported in the literature. Trabelsi and colleagues reported a case of incidental finding in a diabetic patient with external malignant otitis, who underwent bone scintigraphy [9].

Total alkaline phosphatase was assayed in all patients, with two patients showing elevated levels (above 130 IU/L). Elevation of total alkaline phosphatase, a marker of osteoblastic activity, correlates well with the extensive bone remodelling seen in Paget's disease and its extension (in the

absence of liver involvement) [10]. This marker is the most useful for monitoring treatment. Total alkaline phosphatase levels may be normal in monostotic or less extensive forms, or in "extinct" forms. A normal level therefore does not rule out the diagnosis [10, 11-13].

The hyperaemia observed in five (05) patients at the angiographic phase of the affected sites compared with contralateral sites is linked to increased vascular flow during the excessive remodelling found in the affected areas in Paget's disease [8].

The polyostotic form remains frequent (80% in our sample), as described by Stuart and colleagues [5] and Corral-Gudino and colleagues [14] in their respective studies.

A predilection for lesions in the pelvis and femur was found in our study. Other authors have found the same sites of predilection. Stuart et al [5] and Corral-Gudino et al [14] found bone lesions with a predilection for the pelvis and femur.

The multiple bone hyper-fixations found are the consequences of the increased and excessive activity of bone remodelling during Paget's disease. These findings have already been reported by some authors in their studies [6, 13].

Severe limb deformities and varus curvatures of the long bones of the lower limbs have been described in the literature [1, 8].

Bone scintigraphy has made it possible to localize bone damage and characterize disease extension, with a favourable dosimetric ratio [2, 9, 15]. Audran M et al [1] have also reported that bone scintigraphy is an irreplaceable examination in the initial evaluation and follow-up of Paget's disease. Stuart and colleagues [5] add that it is widely regarded as a valuable medical imaging technique for the diagnosis and follow-up of Paget's disease.

5. Conclusion

Paget's disease is a chronic disorder of the adult skeleton, in which the normal matrix is replaced by hypertrophied, less resistant bone. It can lead to progressive bone pain and deformity.

Bone scintigraphy is a fundamental examination for establishing establish a map of the disease's localization at the time of initial management. This examination is more sensitive than radiology, particularly for flat bones.

Whole-body bone scintigraphy plays a key role in the diagnosis of Paget's disease. It is widely regarded as a valuable technique for diagnosis and assessment of disease extent, with a favorable dosimetric ratio. It thus enables early diagnosis and better management of the disease. Its low specificity, however, suggests the need to improve nuclear imaging facilities in Senegal with SPECT/CT and PET/CT.)

Abbreviations

MHBP: Methylene-Hydroxide Bisphosphonate
IPGH: Idrissa Pouye General Hospital
SPECT: Single Photonic Emission Computer Tomography
^{99m}Tc: Metastable Technetium 99

ORCID

0009-0004-4143-585x (El Hadji Amadou Lamine Bathily)

Conflicts of Interest

The authors declare no conflicts of interest.

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