ISA Journal of Medical Sciences (ISAJMS)

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Volume 2, Issue 6, Nov-Dec, 2025 ISSN: 3049-1746

Asymptomatic Diffuse Idiopathic Skeletal Hyperostosis: The Contribution of SPECT/CT to Establishing s Positive Diagnosis

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Received: 21.11.2025 | **Accepted:** 27.11.2025 | **Published:** 03.12.2025

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DOI: *10.5281/zenodo.17805402*

Abstract **Case Studies**

Diffuse idiopathic skeletal hyperostosis (DISH) is a non-inflammatory condition characterized by progressive ossification of the entheses, often asymptomatic and underdiagnosed. We report the case of a 56-year-old female patient followed for a left mandibular carcinoma, with no spinal pain. Whole-body bone scintigraphy, complemented by SPECT/CT of the thoracolumbar spine, revealed a heterogeneous right lateral paravertebral uptake from D5 to D12, corresponding to multilevel anterolateral hyperostosis with cortical thickening and enveloping ligamentous ossification, characteristic of DISH. Additional areas of increased uptake were identified at the mandibular and costal levels, consistent with suspicious neoplastic lesions. This case illustrates the major value of SPECT/CT in confirming the diagnosis of DISH, even when asymptomatic, and in distinguishing degenerative or tumoral abnormalities. Functional imaging coupled with morphological imaging enables precise characterization of enthesopathic abnormalities, facilitates clinical monitoring, and optimizes patient management, thereby reducing the risk of misdiagnosis.

Keywords: DISH, vertebral hyperostosis, bone scintigraphy.

Introduction:

Diffuse idiopathic skeletal hyperostosis (DISH), also known as Forestier's disease, is a non-inflammatory skeletal condition characterized by progressive ossification of the entheses, particularly along the anterior longitudinal ligament of the spine, as well as extraspinal tendon and ligament insertions [1]. Initially described by Forestier and Rotes-Querol in the 1950s under the name of senile ankylosing hyperostosis, this condition has since been widely recognized as a distinct metabolic bone disorder [2].

From an epidemiological standpoint, the prevalence of DISH varies considerably depending on the population and imaging modalities used: studies employing whole-spine computed tomography (CT) have reported prevalences of up to 19% in adult subjects [3]. This variability highlights the fact that DISH is likely underdiagnosed when assessment is limited to conventional radiographs, particularly since the disease is often asymptomatic [4].

Most commonly used diagnostic criteria rely on radiographic findings, notably the criteria established by Resnick and Niwayama, which require the presence of continuous ossifications along at least four contiguous vertebral bodies, preservation of the intervertebral disc space, and absence of inflammatory involvement of the posterior or sacroiliac joints [1]. However, these criteria largely describe an already advanced form of the disease and do not always allow detection of early or subclinical stages [5].

In this context, hybrid SPECT/CT imaging

represents an increasingly relevant tool. Indeed, studies have shown that bone regions affected by DISH may demonstrate delayed increased uptake on SPECT/CT bone scans, which can help differentiate these lesions from other abnormalities such as metastases or arthrosic osteophytes. Moreover, a recently reported clinical case in a young adult showed that SPECT/CT enabled the diagnosis of an early form of DISH, before the full development of the classical radiographic morphological signs [6]. A recent review of the imaging features of DISH emphasized that combining CT with scintigraphy can reveal enthesopathic abnormalities not visible on standard radiographs and contribute to a better understanding of the disease's pathogenesis [7].

Thus, in an asymptomatic form of diffuse hyperostosis, SPECT/CT may play a key role in establishing a positive diagnosis by revealing bone metabolic activity and osteogenic bridging before morphological manifestations are fully mature. This contribution may not only improve early detection but also refine patient stratification according to their risk of progression and guide clinical follow-up.

CASE REPORT:

We report the case of a 56-year-old female patient followed for a left mandibular epithelial–myoepithelial carcinoma. She presented with no spinal pain or stiffness, and the examination was performed as part of the staging workup of her disease. She was referred to the Nuclear Medicine Department of Mohammed VI University Hospital Center in Marrakech. The collected data included age, sex, oncologic history, possible clinical symptoms, as well as the results of scintigraphic and morphological assessments. Bone scintigraphy was performed on a Symbia Pro.specta X3 (Siemens) SPECT-CT gamma camera, including whole-body planar acquisitions followed by SPECT/CT of the skull and thoracolumbar spine, two hours after intravenous injection of 740 MBq (18 mCi) of 99mTc-MDP.

Planar acquisitions (Figure 1) revealed heterogeneous uptake of the dorsal spine, more marked on the right lateral paravertebral region, with a focal hotspot at D12. In addition, several other areas of increased uptake were identified: an intense focus at the left mandibular angle and a moderate focus at the posterior arch of the 7th left rib. Degenerative-appearing uptakes were also observed in the shoulders, knees and ankles. Renal and urinary tract activity was moderate with physiological distribution, and the radiotracer distribution was symmetric and homogeneous throughout the remainder of the skeleton.

SPECT/CT acquisition (Figure 2) demonstrated heterogeneous right lateral paravertebral hyperuptake extending from D5 to D12, corresponding to multilevel anterolateral hyperostosis with cortical thickening and enveloping ligamentous ossification, typical features of Forestier's disease. This correlation between morphological abnormalities and scintigraphic hyperuptake enabled the positive diagnosis of DISH, illustrating the value of SPECT/CT in detecting this asymptomatic diffuse vertebral hyperostosis. In addition, other foci of increased uptake were identified: an intense focus at the left mandibular angle corresponding to a Lodwick type II osteolytic lesion, a moderate focus on the posterior arch of the 7th left rib corresponding to a sclerotic lesion suspicious for secondary localization, as well as two sclerotic abnormalities on the anterior arch of the 4th left rib and the right iliac wing with no scintigraphic expression.

Thus, SPECT/CT allowed the specific identification and confirmation of Forestier's disease while discriminating suspicious foci of neoplastic or degenerative origin. This case highlights the major value of combined functional and morphological imaging in the positive diagnosis of this asymptomatic diffuse skeletal hyperostosis.

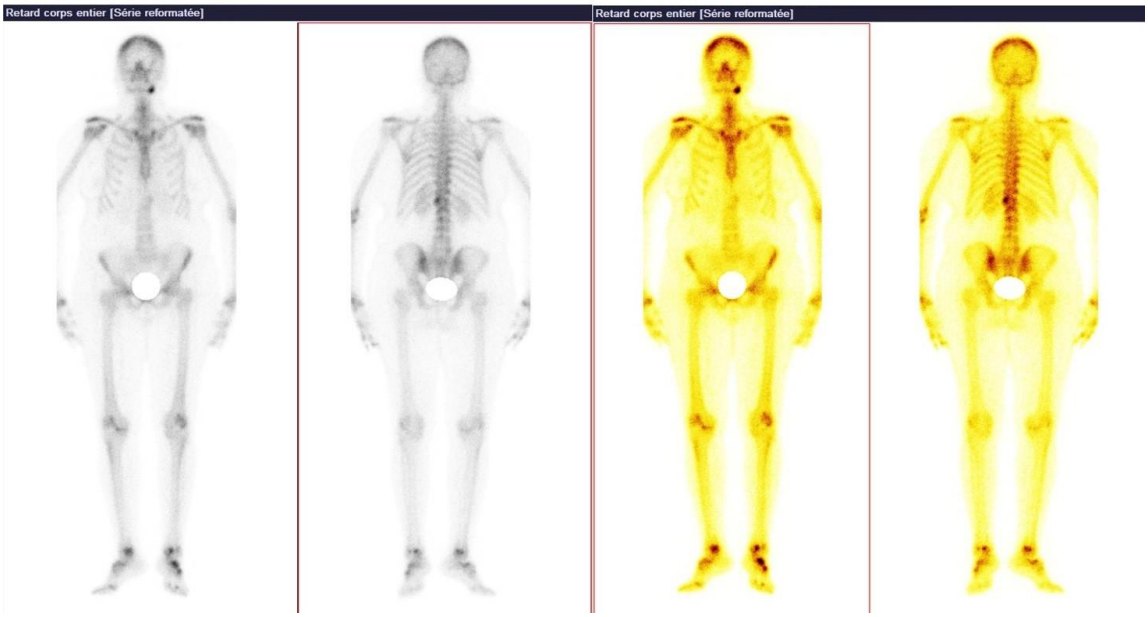

Figure 1: Whole-body planar imaging in anterior and posterior views

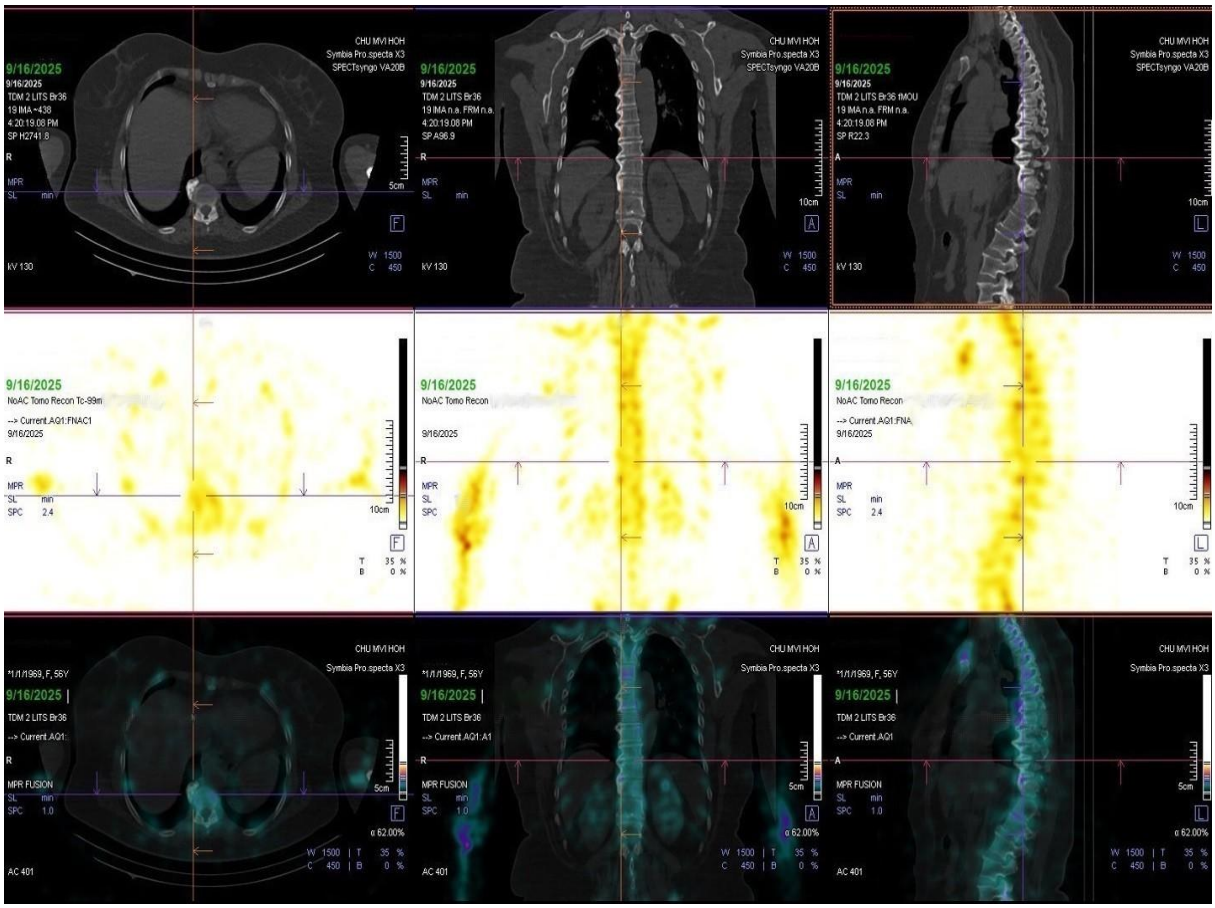

Figure 2: SPECT/CT acquisition of the thoracolumbar spine.

Discussion:

The observation of this case highlights the clinical and diagnostic significance of diffuse idiopathic skeletal hyperostosis (DISH) as a major entity, even in the absence of spinal symptoms. DISH is a non-inflammatory condition characterized by progressive ossification of the entheses, particularly along the anterior longitudinal ligament of the spine [8].

The prevalence of DISH is likely underestimated when relying solely on radiographs, as studies using computed tomography (CT) show higher rates. For example, a study of 1,815 whole-spine CT scans reported a prevalence of 19.1% [9].

Similarly, in a cohort of 1,479 subjects evaluated by spinal CT, the prevalence was 19.5%, with ossifications primarily located in the thoracic region, notably T8–T10 [10].

A systematic and meta-analytic review estimated an overall prevalence of approximately 11.9% in the general population, with higher rates among clinical patients [11].

These data emphasize that DISH is far from rare, especially in older adults, but may go unnoticed when asymptomatic.

Everal factors are associated with DISH: advanced age, male sex, obesity, diabetes, and cardiovascular comorbidities such as atherosclerosis [12]. These associations suggest that metabolic and vascular factors may play a role in the disease's pathogenesis.

The exact pathophysiology of DISH remains poorly understood. Enthesial bone formation appears to be influenced by a combination of mechanical, metabolic, vascular, and possibly genetic factors [12]. Enthesial fibroblasts, chondrocytes, and the extracellular matrix are thought to be activated in response to these stimuli, promoting ectopic ossification.

Although DISH is often asymptomatic, as in our patient, it is not necessarily clinically benign. Spinal rigidity may occur, leading to limitations in mobility, even if subclinical [13]. The risk of vertebral fracture is increased, as spines ankylosed by ossification are more fragile and prone to unstable fractures [14]. Rare but clinically significant extraskeletal manifestations can occur, for example, large anterior cervical osteophytes may induce dysphagia, airway obstruction, or neurological disorders [15]. Associations with other ligamentous ossifications, such as ossification of the posterior longitudinal ligament or ligamentum flavum, have also been reported [16].

In our case, the use of SPECT/CT enabled unequivocal confirmation of DISH diagnosis, due to the correlation between scintigraphic hyperuptake and morphological bone changes. This approach offers several advantages, such as the sensitivity of SPECT, which can detect bone metabolic activity even in areas long considered clinically silent, while the specificity of CT allows precise characterization of bone architecture, including ossifications, densification, and bony bridging, and differentiates DISH from other lesions such as metastases or arthritic osteophytes. This level of detail is particularly crucial, as it helps prevent misdiagnoses, including confusion with neoplastic sites, and effectively guides patient management and clinical follow-up.

In addition to DISH, other foci of increased uptake identified in our case were of neoplastic nature. The coexistence of these foci with asymptomatic DISH underscores the importance of a rigorous differential diagnosis. SPECT/CT plays a key role in this distinction, helping prioritize monitoring of suspicious areas and guiding potential further investigations.

Our case emphasizes the value of considering Forestier's disease as a central diagnosis, even in the absence of spinal symptoms, when scintigraphic hyperuptakes are detected. The use of SPECT/CT not only confirms the diagnosis but also differentiates suspicious lesions and guides follow-up. Finally, this highlights the need to raise awareness of this very common, often asymptomatic condition, which may nonetheless carry significant clinical complications.

CONCLUSION

Diffuse idiopathic skeletal hyperostosis

(DISH), or Forestier's disease, is a common yet often underrecognized condition that can be asymptomatic despite significant radiological abnormalities. Its diagnosis relies on the identification of characteristic enthesopathic ossifications and assessment of vertebral morphology, and it can be confirmed early through functional imaging combined with morphological evaluation, such as SPECT/CT. This approach provides increased sensitivity for detecting bone activity, as well as high specificity for distinguishing DISH from tumoral or degenerative lesions, thereby improving clinical management and patient follow-up. Systematic recognition of this condition and the combined use of SPECT and CT thus allow a better understanding of its distribution, progression, and potential impact, while guiding surveillance and preventing misdiagnosis, emphasizing the importance of routinely acknowledging this entity in medical practice.

Acknowledgements: The authors wish to thank all colleagues who supported this study.

Authorship Contributions: Medical Practices**:** S.E.A., H.A., M.A.B., A.M., Concept: S.E.A., Design: S.E.A., Data Collection or Processing: S.E.A., Analysis or Interpretation: S.E.A., Literature Search: S.E.A., Writing: S.E.A.,

Funding: This research received no specific grant from any funding agency in the public, commercial or not- for-profit sectors.

Declaration

Conflict of interest: All the authors (Saad Eddine Abaid, Hamza Alaoui, Mohammed Aziz Bsiss, Aboubaker Matrane) declare that there is no confict of interest that could be perceived as prejudicing the impartiality of the research reported.

Informed consent: Written informed consent was obtained prospectively from the patient to use their clinical data.

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