

**ADDED VALUE OF HYBRID IMAGING IN THE DIAGNOSIS AND MANAGEMENT OF  
OSTEOID OSTEOMA: CASE REPORTS**

Sanae El Mselmi\*, Fabrice Fokoué, Nadia Ismaili Alaoui

Department of Nuclear Medicine, Oncology Hospital, Hassan II University Hospital Center, Fez, Morocco.

**\*Corresponding Author: Sanae El Mselmi**

Department of Nuclear Medicine, Oncology Hospital, Hassan II University Hospital Center, Fez, Morocco.

Article Received on 10/03/2022

Article Revised on 30/03/2022

Article Accepted on 20/04/2022

**ABSTRACT**

Osteoid osteoma is a painful, benign, osteoblastic lesion that occurs in younger patients and affects the extremities or the axial skeleton. Osteoid osteoma has previously been diagnosed using plain radiograph imaging. However, diagnosis of osteoid osteoma may be delayed due to ambiguities on plain radiograph images; despite the increasing use of magnetic resonance imaging (MRI), this type of misdiagnosis is not uncommon. Through two case reports, the aim of the present study was to evaluate the effectiveness of radionuclide imaging scans with SPECT/CT for the diagnosis of osteoid osteoma, as this form of imaging was proposed to be a more sensitive test.

**INTRODUCTION**

Osteoid osteoma is an osteoblastic bone tumor which accounts for 10% of all benign bone tumours and 4% of all primary bone tumours. Osteoid osteoma is the third most common benign bone tumour after enchondroma and nonossifying fibroma.<sup>[1-2]</sup> It generally affects younger, predominantly male patients with a peak incidence in the second decade of life. Bergstrand was the first to describe osteoid osteoma in 1935, and Jaffe subsequently characterized osteoid osteoma as a distinct clinical entity.<sup>[3-4]</sup> This tumour is described as a well-demarcated osteoblastic mass, called a nidus, surrounded by extensive formation of compact bone.

Osteoid osteomas may occur anywhere within the axial or appendicular skeleton, with the majority (>50%) occurring in lower extremity locations such as the femur or tibia.<sup>[2]</sup> Within the bone, osteoid osteomas preferentially involve the cortex of long bones, usually in the diaphysis or metadiaphysis.<sup>[2]</sup> The classic clinical presentation includes local bone pain, often worse at night and relieved by nonsteroidal anti-inflammatory drugs (NSAIDs). In the majority of osteoid osteoma cases, typical radiographic features demonstrate a sclerotic cortical lesion and contain a small lucency that represents a nidus.<sup>[5-6]</sup>

The present study aims to investigate the potential role of hybrid imaging bone scan for the diagnosis of osteoid osteoma, as radionuclide imaging has been reported to be a more sensitive diagnostic modality in osteoid osteoma.

**Case reports****Case 1:**

An 8-year-old boy with a limp with right hip pain evolving for six months with no other associated signs. The clinical examination reveals a painful right lower limb mobility. The radiograph shows a small metaphysical subperiosteal nidus and thickening of the medial cortex face of the right femur (Fig. 1). A bone scintigraphy was performed using <sup>99m</sup>Tc-methylene diphosphonate (MDP) and the bone scan image revealed increased focal uptake in the posteromedial aspect of the right femur (Fig. 2). The SPECT CT images (Fig. 3) show a subperiosteal nidus and cortical thickening. The diagnosis of osteoid osteoma in the right femur was confirmed.

The child benefited from a tumor resection. Anatomopathological examination confirmed the diagnosis of osteoid osteoma.



Fig. 1: The radiograph shows a small subperiosteal nidus (Arrow) and Thickening of the medial cortex.

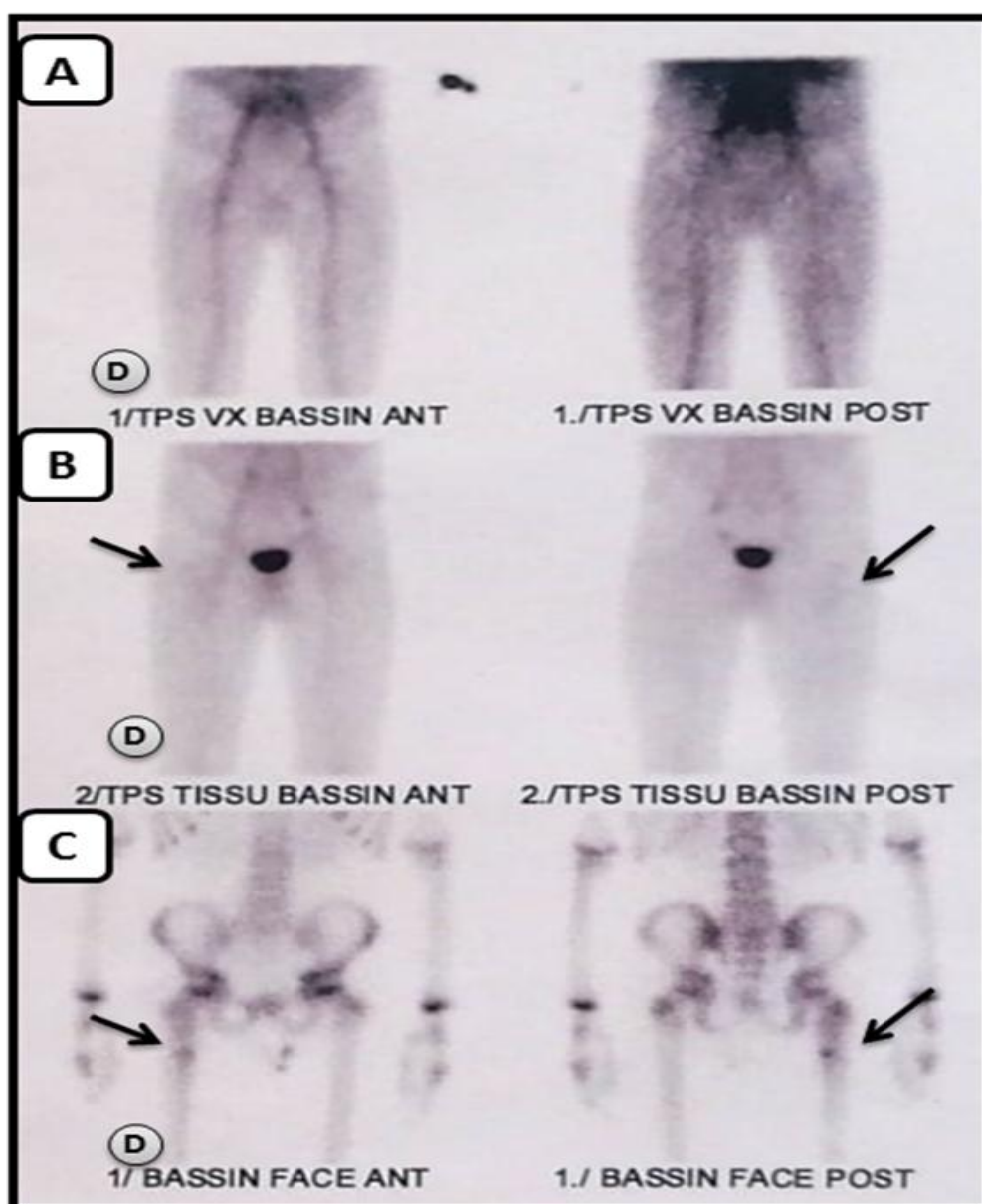
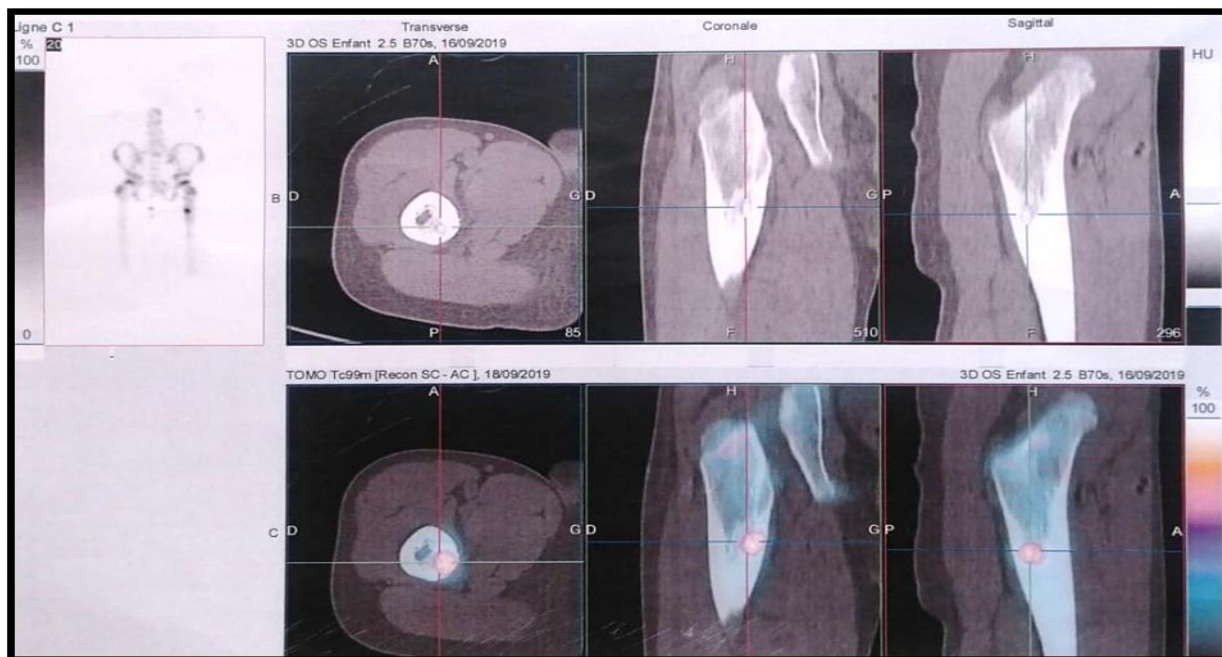


Fig. 2: A/B: The early phase planar bone scan images show slightly increased uptake (arrow) in the posteromedial aspect of the right femur. C: The late phase bone scan images show markedly increased uptake (arrows) in the posteromedial right femur.

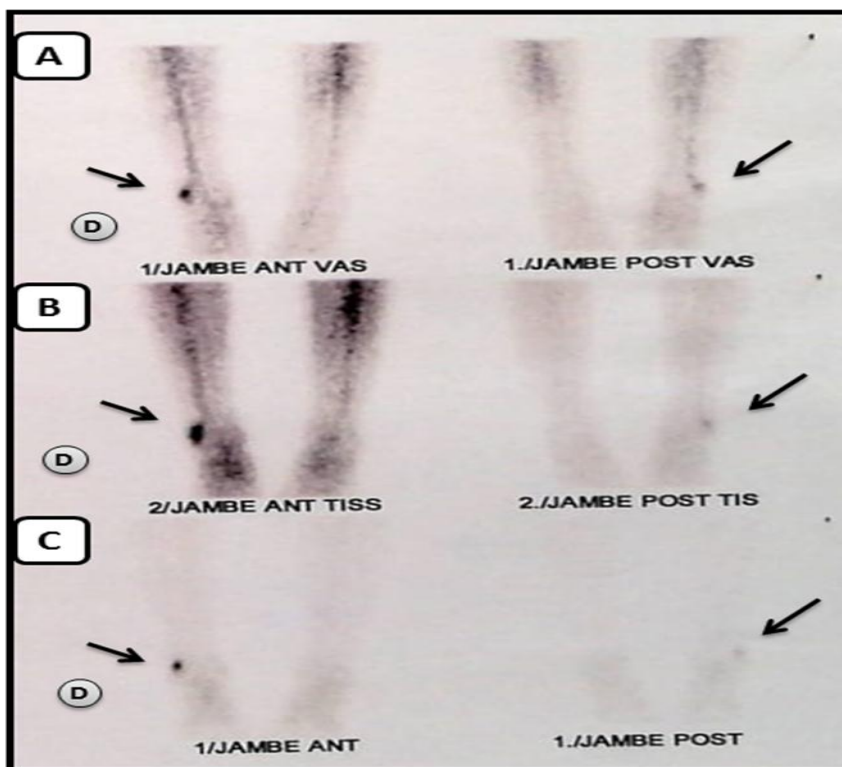


**Fig. 3:** The SPECT CT images show a subperiosteal nidus (Arrows) and cortical thickening.

**Case 2:**

A 15-year-old girl presented with right ankle pain, which worsened at night. The patient's symptoms had lasted for one year and were treated as an ankle sprain at another hospital based on the results of plain radiographs, which revealed no specific bony abnormalities. A bone scintigraphy (Fig. 4) and SPECT CT (Fig. 5) scans were

performed. The early phase planar bone scan images show markedly increased uptake in the anterolateral aspect of the right ankle. The late phase bone scan images show markedly increased uptake in the anterolateral right ankle. The SPECT CT images show a nidus in the peroneal malleolus.



**Fig. 4:** A: The early phase planar bone scan images show markedly increased uptake in the anterolateral aspect of the right ankle. B: The late phase bone scan images show markedly increased uptake in the anterolateral right ankle.



**Fig. 5: The SPECT CT images show a nidus in the peroneal malleolus.**

## DISCUSSION

Osteoid osteoma is a type of benign bone-forming tumor which is characterized by a well-demarcated osteoblastic mass, called a nidus, surrounded by a distinct zone of reactive bone sclerosis.<sup>[8]</sup> The pathogenesis of osteoid osteoma remains unclear; some authors suggest that it is a true benign osteoblastic neoplasm, while others believe it may represent unusual healing or an inflammatory process.<sup>[9-10]</sup> This tumor accounts for 10-12% of all benign bone tumours (2) and generally affects younger, predominantly male.<sup>[14]</sup>

Osteoid osteomas may occur anywhere within the axial or appendicular skeleton, with the majority (>50%) occurring in lower extremity locations such as the femur or tibia.<sup>[2,16]</sup>

Two thirds of the femoral lesions are situated in the intertrochanteric or intracapsular regions of the hip. The humerus is another common location for osteoid osteoma. Within the bone, osteoid osteomas preferentially involve the cortex of long bones, usually in the diaphysis or metadiaphysis<sup>[2,17]</sup> Approximately 10% to 20% of osteoid osteomas occur in the spine, most commonly in the lumbar spine with a predilection for the posterior elements. The vertebral body is often spared, being involved in only 10% of spinal lesions.<sup>[11,18,19]</sup> Uncommon locations for osteoid osteoma include the skull, clavicle, shoulder, hand, pelvis, ankle and foot.<sup>[11,14,15]</sup>

The classic clinical presentation includes local pain, often worse at night and relieved by nonsteroidal anti-inflammatory drugs following a local production of prostaglandins at very high levels.<sup>[12,20,21]</sup> This pain is often referred to the nearest joint when the tumor is located in the proximity of a joint, which physicians may confuse with arthritic pain.<sup>[13,22]</sup> Other clinical features associated with osteoid osteoma depend on the specific location of the lesion, and include swelling, limp, painful scoliosis, growth disturbance, joint stiffness and contracture.<sup>[15]</sup>

The radiographic appearance of osteoid osteoma depends on its location within the involved bone. The most common appearance is a cortical-based lucency measuring less than 2 cm, with central calcification seen within a radiolucent nidus, especially in larger lesions.<sup>[1,15,23]</sup> The typical radiographic features of osteoid osteoma are not always distinguishable. Intracortical lesions of long bones produce extensive fusiform thickening of the cortex with dense radiopacity, which may obscure the nidus of osteoid osteoma.<sup>[1,24]</sup> The degree of sclerosis surrounding the nidus is less in epiphyseal and metaphyseal lesions than in diaphyseal lesions; medullary lesions also exhibit less sclerosis than their intracortical counterparts. Subperiosteal lesions produce minimal sclerosis and may appear as soft-tissue lesions adjacent to the affected bone, which reveals irregular bony resorption.<sup>[17]</sup> According to Swee et al., plain radiograph images and clinical history were sufficient for the accurate diagnosis of osteoid osteoma in 75% of cases.<sup>[12]</sup> Jordan et al., in a meta-analysis of

223 patients with proven osteoid osteoma, found a detection rate of only 66% by plain radiography.<sup>[26]</sup> Plain radiography might fail to depict an osteoid osteoma in complex anatomical areas such as the spine, pelvis, skull or foot, where superimposed bony structures can obscure the lesion.<sup>[12,27,28,29]</sup>

CT is far superior to plain radiography for the detection and characterization of osteoid osteoma with high sensitivity compared to plain radiography.<sup>[16,30,31]</sup>

The typical lesion is seen as a well-defined round or oval focal area of soft-tissue attenuation, less dense than, and surrounded by, variable amounts of osteosclerosis. CT is especially useful for the detection of spinal osteoid osteomas, which are seen as low-density lesions in the posterior elements of the vertebral column associated with reactive sclerosis of the ipsilateral pedicle, lamina or transverse process. Several authors have reported a 96-100% detection rate of osteoid osteoma using CT.<sup>[26,28,29]</sup>

Although many studies have shown that CT is superior to MRI for detecting and characterizing osteoid osteoma.<sup>[25,32]</sup> The MRI appearance of osteoid osteoma is variable, commonly exhibiting low to intermediate T1-weighted signal and heterogeneous high signal with T2-weighted and STIR sequences.<sup>[16,25,33]</sup> Central nidus calcifications show low signal with both T1-weighted and T2-weighted sequences.<sup>[11]</sup> Most osteoid osteomas enhance diffusely after gadolinium administration as a result of their intrinsic vascularity. Rim enhancement may be heterogeneous. Perilesional sclerosis is seen as fusiform low signal with both T1-weighted and T2-weighted sequences. MRI typically shows intense surrounding bone marrow and soft tissue oedema. However, some authors have reported a 35% risk of misdiagnosis using MRI as the first choice technique for the diagnosis of osteoid osteoma,<sup>[25,26,33]</sup> especially in small tumor.

Bone scintigraphy with 99mTc-labelled bisphosphonates has been used for decades for the diagnosis of osteoid osteoma, with a reported sensitivity of nearly 100%.<sup>[34,35]</sup>

The classic bone scintigraphic finding is the double density sign, where a central focus of very high activity corresponding to the nidus of the osteoid osteoma is surrounded by a larger area of less intense radiopharmaceutical uptake, representing the host bone tumour response.<sup>[15,23,36,37,38]</sup> This sign is very specific for osteoid osteoma in the appendicular skeleton; however, it is less frequently seen in spinal lesions because of less reactive osteosclerosis in the vertebrae.<sup>[36,39]</sup> The presence of the pathognomonic double density sign allows differentiation of osteoid osteoma from Brodie's abscess, stress reaction and metastasis.<sup>[1,2]</sup> In comparison with planar imaging, single photon emission computed tomography (SPECT) imaging with three dimensional reconstruction techniques has better spatial resolution and is therefore able to detect smaller lesions.<sup>[40,41]</sup> To

further improve the detection and characterization of osteoid osteoma, SPECT examinations are typically followed by a coregistered or stand-alone CT examination. The fused functional and structural information provided by most current SPECT/CT scanners has led to this modality becoming the one-stop imaging tool that is able to diagnose osteoid osteoma with the highest accuracy.<sup>[42,43]</sup> In a retrospective study of 31 patients examined for suspected osteoid osteoma, Sharma et al. found significantly higher sensitivity, specificity and accuracy with SPECT/CT (all 100%) than with CT (77.8%, 92.3% and 83.8%, respectively) or planar bone scintigraphy (100%, 38.4% and 74.1%, respectively).<sup>[44]</sup>

## CONCLUSION

Osteoid osteomas are benign, often painful bone tumours that are frequently challenging to detect and characterize on plain radiography and even MRI, especially in complex anatomical regions such as the spine, pelvis, wrist and foot. Hybrid imaging (SPECT/CT with 99mTc-labelled bisphosphonates or PET/CT with 18F-NaF) is highly accurate and provides all the necessary functional information for diagnosis, as well as morphological information to guide treatment.

## REFERENCE

1. Chai JW, Hong SH, Choi JY, Koh YH, Lee JW, Choi JA, et al. Radiologic diagnosis of osteoid osteoma: from simple to challenging findings. *Radiographics*, 2010; 30(3): 737-49.
2. Kransdorf MJ, Stull MA, Gilkey FW, Moser RP Jr. Osteoid osteoma. *Radiographics*, 1991; 11(4): 671-96.
3. Bergstrand H. Uber eine eigenartige, wahrscheinlich bisher nicht beschriebene osteoblastische krankheit in den langen knochen der hand und des fusses. *Acta Radiol*, 1930; 11: 596-613.
4. Jaffe H. Osteoid osteoma: a benign osteoblastic tumor composed of osteoid and atypical bone. *Arch Surg*, 1935; 31: 709-28.
5. Schajowicz F and Lemos C: Osteoid osteoma and osteoblastoma. Closely related entities of osteoblastic derivation. *Acta Orthop Scand*, 1970; 41: 272-291.
6. Sim FH, Dahlin CD and Beabout JW: Osteoid-osteoma: Diagnostic problems. *J Bone Joint Surg Am*, 1975; 57: 154-159.
7. Becce F, Theumann N, Rochette A, Larousserie F, Campagna R, Cherix S et al. Osteoid osteoma and osteoid osteomamimicking lesions: biopsy findings, distinctive MDCT features and treatment by radiofrequency ablation. *Eur Radiol*, 2010; 20(10): 2439-46.
8. Greenspan A: Benign bone-forming lesions: Osteoma, osteoid osteoma and osteoblastoma.. Clinical, imaging, pathologic and differential considerations. *Skeletal Radiol*, 1993; 22: 485 - 500.

9. JS Kneisl, Simon MA. Medical management compared with operative treatment for osteoid-osteoma. *J Bone Joint Surg Am*, 1992; 74: 179-85.
10. Lee EH, Shafi M, Hui JH. Osteoid osteoma: a current review. *J Pediatr Orthop*, 2006; 26(5): 695-700.
11. Sabanas AO, Bickel WH and Moe JH: Natural history of osteoid osteoma of the spine; review of the literature and report of three cases. *Am J Surg*, 1956; 91: 880-889.
12. Swee RG, McLeod RA, Beabout JW. Osteoid osteoma. Detection, diagnosis, and localization. *Radiology*, 1979; 130: 117-23.
13. Georgoulis AD, Papageorgiou CD, Moebius UG, Rossis J, Papadnikolakis A and Soucacos PN: The diagnosis dilemma created by osteoid osteoma that presents as knee pain. *Arthroscopy*, 2002; 18: 32-37.
14. Chicoisne MP, Assoun J, Giron J, Railhac JJ. Osteoid osteoma. *EMC Radiology and Musculoskeletal Medical Imaging - Neurological - Maxillofaciale*, 1993. [31-482-A-10].
15. Iyer RS, Chapman T, Chew FS. Pediatric bone imaging: diagnostic imaging of osteoid osteoma. *AJR Am J Roentgenol*, 2012; 198: 1039-52.
16. Papathanassiou ZG, Megas P, Petsas T, Papachristou DJ, Nilas J, Siablis D. Osteoid osteoma: diagnosis and treatment. *Orthopedics*, 2008; 31(11): 1118.
17. Edeiken J, DePalma AF, Hodes PJ. Osteoid osteoma. (Roentgenographic emphasis). *Clin Orthop Relat Res*, 1966; 49: 201-6.
18. Graham GN, Browne H. Primary bony tumors of the pediatric spine. *Yale J Biol Med.*, 2001; 74(1): 1-8.
19. Healey JH, Ghelman B. Osteoid osteoma and osteoblastoma. Current concepts and recent advances. *Clin Orthop Relat Res*, 1986; 204: 76-85.
20. Greco F, Tamburrelli F, Ciabattini G. Prostaglandins in osteoid osteoma. *Int Orthop*, 1991; 15: 35-7.
21. Mungo DV, Zhang X, O'Keefe RJ. COX-1 and COX-2 expression in osteoid osteomas. *J Orthop Res*, 2002; 20: 159-62.
22. Greenspan A, Jundt G and Remagen W (eds): eds Bone forming (osteogenic) osteogenic lesions. In: *Differential Diagnosis in Orthopaedic Oncology*. 2nd edition. Lippincott Williams and Wilkins, Philadelphia, PA, 2006; 40-157.
23. Allen SD, Saifuddin A. Imaging of intra-articular osteoid osteoma. *Clin Radiol*, 2003; 58: 845-52.
24. Ghanem I. The management of osteoid osteoma: updates and controversies. *Curr Opin Pediatr*, 2006; 18(1): 36-41.
25. Assoun J, Richardi G, Railhac JJ, Baunin C, Fajadet P, Giron J, et al. Osteoid osteoma: MR imaging versus CT. *Radiology*, 1994; 191(1): 217-23.
26. Jordan RW, Koc T, Chapman AW, Taylor HP. Osteoid osteoma of the foot and ankle - a systematic review. *Foot Ankle Surg*, 2015; 21 (4): 228-34.
27. Helms CA, Hattner RS and Vogler JB III: Osteoid osteoma: Radionuclide diagnosis. *Radiology*, 1984; 151: 779-784.
28. JL Gamba, Martinez S, Apple J, Harrelson JM, Nunley JA. Computed tomography of axial skeletal osteoid osteomas. *AJR Am J Roentgenol*, 1984; 142(4): 769-72.
29. Harish S, Saifuddin A. Imaging features of spinal osteoid osteoma with emphasis on MRI findings. *Eur Radiol*, 2005; 15(12): 2396-403.
30. Levine E, Neff JR. Dynamic computed tomography scanning of benign bone lesions: preliminary results. *Skelet Radiol*, 1983; 9(4): 238-45.
31. McGrath BE, Bush CH, Nelson TE, Scarborough MT. Evaluation of suspected osteoid osteoma. *Clin Orthop Relat Res*, 1996; 327: 247-52.
32. Zanetti M, Eberhard SM, Exner GU, von Hochstetter A, Hodler J. Magnetic resonance tomography in osteoid osteoma: more confusion than benefit?. *Praxis (Bern 1994)*, 1997; 86(11): 432-6.
33. Davies M, Cassar-Pullicino VN, Davies AM, McCall IW, Tyrrell PN. The diagnosis accuracy of MR imaging in osteoid osteoma. *Skelet Radiol*, 2002; 31(10): 559-69.
34. Wells RG, Miller JH, Sty JR. Scintigraphic patterns in osteoid osteoma and spondylolysis. *Clin Nucl Med*, 1987; 12(1): 39-44.
35. Park JH, PakhK, KimS, Lee SH, Song SH, JG Choe. Radionuclide imaging in the diagnosis of osteoid osteoma. *Oncol Lett*. 2015; 10(2): 1131-4.
36. Helms CA. Osteoid osteoma. The double density sign. *Clin Orthop Relat Res*, 1987; 222: 167-73.
37. Chrestian P, Vaylet C, Puech B, Nicaud M, Maximin MC. The contribution of the scan in the pathology of the musculoskeletal system of the child and the adolescent. Cross-eyed glances of the clinician and the scintigrapher. *Med Nucl*, 2012; 36: 128-39.
38. Hod N, Fire G, Horne T. A rare case of osteoid osteoma of the coccyx detected on bone scintigraphy. *Clin Nucl Med*, 2006; 31: 550-1.
39. Roach PJ, Connolly LP, Zurakowski D, Treves ST. Osteoid osteoma: comparative utility of high-resolution resolution planar and pinhole magnification scintigraphy. *Pediatr Radiol*, 1996; 26(3): 222-5.
40. Banzo I, Montero A, Uriarte I, Vallina NK, Hernandez A, Guede C, et al. Localization by bone SPET of osteoid osteoma in the vertebral lamina. *Rev Esp Med Nucl*, 1999; 18(1): 47-9.
41. Ryan PJ, Fogelman I. Bone SPECT in osteoid osteoma of the vertebral lamina. *Clin Nucl Med*, 1994; 19 (2): 144-5.
42. Hasegawa BH, Wong KH, Iwata K, Barber WC, Hwang AB, Sakdinawat AE, et al. Dual-modality imaging of cancer with SPECT/CT. *Technol Cancer Res Treat*, 2002; 1(6): 449-58.
43. Squier SB, Lewis JI, Accurso JM, Jain MK. (99m) Tc-methylene diphosphonate single-photon emission

computed tomography/ computed tomography improves the diagnosis accuracy of osteoid osteoma. Indian J Nucl Med, 2016; 31(4): 298-300.

44. Sharma P, Mukherjee A, Karunanithi S, Nadarajah J, Gamanagatti S, Khan SA, et al. 99mTc-methylene diphosphonate SPECT/CT as the one-stop imaging modality for the images of osteoid osteoma.. Nucl Med Commun, 2014; 35 (8): 876-83.