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The evaluation of SPECT/CT bone scintigraphy in metastatic fibrous dysplasia

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► ***Case Report***

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ABSTRACT

Background: Fibrous dysplasia is a rare benign bone disorder characterized by the fibrous tissue containing trabeculae of non-lamellar bone (woven bone) and occupying normal medullary spaces. The uptake of the radiotracer in the affected bone is variable, and specificity is too low to diagnose fibrous dysplasia in the bone scan. ***Materials and Methods:*** We are presenting four cases with monostotic fibrous dysplasia which were detected incidentally on the routine planar bone scan while being investigated for a metastatic cancer work-up. During a cancer work-up at our institution, we find lesions of fibrous dysplasia showing significantly increased uptake on the bone scan which may mimic metastatic bone lesions. ***Results:*** The SPECT/CT increases diagnostic confidence and improves accuracy and specificity of a planar bone scan. These cases were all asymptomatic at the one-year follow-up The subsequent bone scan and radiography studies have revealed no progression of these bone lesions. ***Conclusion:*** The SPECT/CT images increase the diagnostic accuracy of the bone scan, which may avoid unnecessary surgery or overtreatment of fibrous dysplasia as bone metastasis. ***Abbreviation:*** SPECT/CT = single photon emission computed tomography/computed tomography.

Keywords: *Bone scan, bone metastasis, fibrous dysplasia, SPECT/CT.*

INTRODUCTION

Fibrous dysplasia was first described by Lichtenstein in 1938 as a disorder characterized by progressive replacement of normal bone elements by fibrous tissue. Fibrous dysplasia of bone is a rare benign bone disorder affliction accounting for 0.8 to 7% of all bone benign tumors [1-3]. It has poliostotic and monostotic patterns. Monostotic fibrous dysplasia is frequently asymptomatic and is usually discovered incidentally by radiologic imaging performed for other reasons [4]. The tumor may remain undetected throughout the lifetime of the patient. We believe that a significant portion of patients with the disease remain undiagnosed. The radiotracer uptake is variable and specificity is too low to diagnose fibrous dysplasia with a bone scan [5]. However, fibrous dysplasia in some cases, shows significant, focally increased radioactivity on the ^{99m}Tc-MDP bone scan, which can be a difficult differential diagnosis from bone metastasis. It is a challenge and needs to be characterized. The main limitation of the planar bone scan to date has been limited specificity and lack of anatomical data which leads to

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difficulty of accurate localization of the abnormal tracer uptake. This has all changed with the introduction of SPECT/CT. The authors demonstrate the usefulness of SPECT/CT for the differentiated diagnosis of fibrous dysplasia from osseous metastasis in four cases with known cancer, showing hot spots in the routine bone scan. These cases illustrate potential pitfalls of unexpected fibrous dysplasia misdiagnosed as stage-changing bone metastases [6-8]. The SPECT/CT reduces the number of equivocal findings on the planar scan, increases the diagnostic confidence, and improves accuracy and specificity.

Case Reports

This study was conducted retrospectively to analyze the medical records of patients with SPECT/CT bone scintigraphy in the Department of Nuclear Medicine, Kaohsiung Medical University Hospital. The review process was approved by the Institutional Review Board KMUHIRB-20180009.

Case 1

A 43 year-old woman newly diagnosed with left breast cancer, pT1cN0M0, stage 1A. For evaluation of metastatic bone disease, the whole body bone scan was performed at three hours after the injection of 740MBq (20 mCi) ^{99m}Tc methylene diphosphonate (MDP). No definite abnormal bone lesion was shown except a hot spot in the anterior aspect of the right first rib (figure 1-A). The SPECT/CT study was then performed while the patient remained on the scanning bed, and typical radiolucent and sclerotic change with bone expansion in fibrous dysplasia over the right first rib was noted on the reference CT images (figure 1-B). This finding was not indicated on the first interpretation of a previous chest plain film, before the result of the bone scan was noted. (figure 1-C).

Case 2

A 67 year-old man newly diagnosed with prostate cancer, adenocarcinoma, Gleason score 3+4=7, pT3a N0, MRI TNMStage: T2aN0Mb. The planar whole body images showed intense radiotracer accumulation in the left superior orbital margin and adjacent left frontal skull (figure 2-A). A head plain film, CT or MRI of this patient was not available at that time. The subsequent SPECT/CT study showed typical radiolucent areas with various ossification, cystic formation and bone expansion in fibrous dysplasia involving the left frontal bone, adjacent left frontal, ethmoid and sphenoid sinuses on the reference CT images (figure 2-B).

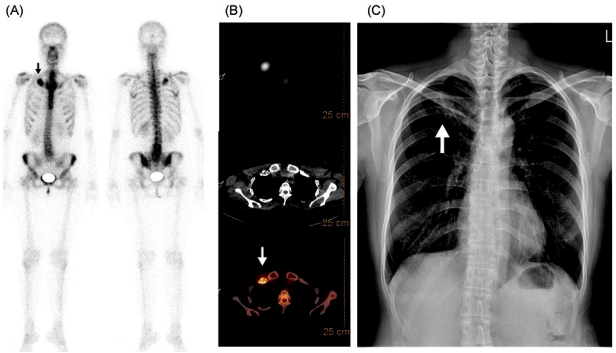

Figure 1. (A) The planar bone scan: a hot spot in anterior aspect of right 1st rib. **(B)** The SPECT/CT study: typical radiolucent and sclerotic change with bone expansion in fibrous dysplasia over right 1st rib was noted on the reference CT images. **(C)** This finding was not pointed out on the first interpretation of a previous chest plain film.

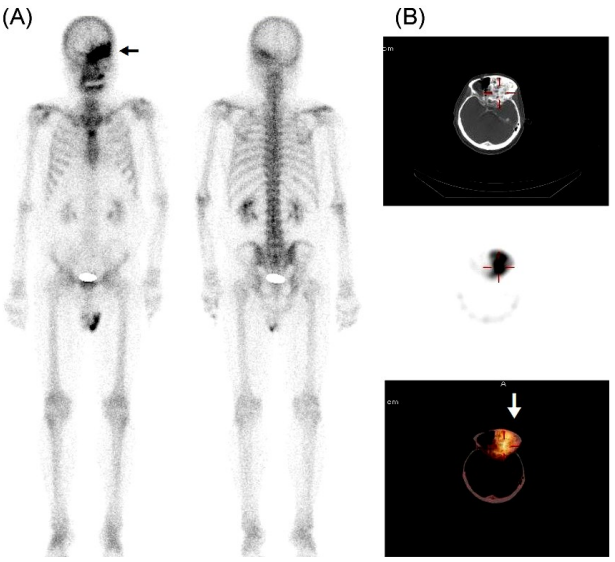

Figure 2. (A) The planar bone scan: intense radiotracer accumulation in left superior orbital margin and adjacent left frontal skull. **(B)** The SPECT/CT study: typical radiolucent areas with various ossification, cystic formation and bone expansion involving left frontal bone, adjacent left frontal, ethmoid and sphenoid sinuses on the reference CT images.

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Case 3

A 62 year-old woman with newly diagnosed right renal pelvis infiltrating urothelial carcinoma, high grade, pT4. The planar whole body bone scan showed focal intense uptake in the proximal portion of the right humeral shaft (figure 3-A). The subsequent SPECT/CT study shows irregular areas with various ossification and cystic formation in the right proximal humeral cortex and adjacent marrow space on the reference CT images (figure 3-B). This finding was not noted on the first interpretation of a previous chest plain film before the bone scan was resulted. (figure 3-C).

Two weeks later, a subsequent imaging survey was performed including maximum intensity projection ^{18}F-fluorodeoxyglucose positron emission tomography and computed tomography (^{18}F-FDG PET/CT). No obvious ^{18}F-FDG uptake in this area (figure 3-D) was demonstrated.

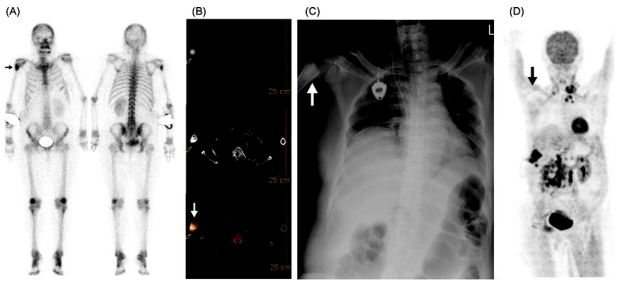

Figure 3. (A) The planar bone scan: focal intense uptake in proximal portion of right humeral shaft. **(B)** The SPECT/CT study: irregular areas with various ossification and cystic formation in right proximal humeral cortex and adjacent marrow space on the reference CT images. **(C)** This finding was not pointed out on the first interpretation of a previous chest plain film. (D) Maximum intensity projection 18F-FDG PET/CT images: no obvious 18F-FDG uptake in this area.

Case 4

A 63 year-old man with newly diagnosed lung cancer, adenocarcinoma, T2aN3M1c, stage IVB. The planar whole body images showed a hot spot in a lower portion of the right ilium (figure 4-A). The SPECT/CT images showed a typical appearance with various ossification and cystic formation in fibrous dysplasia over the right ilium on the reference CT images (figure 4-B). This finding was not pointed out on the first interpretation of previous lumbar spine radiography performed 3 years ago from another hospital (figure 4-C).

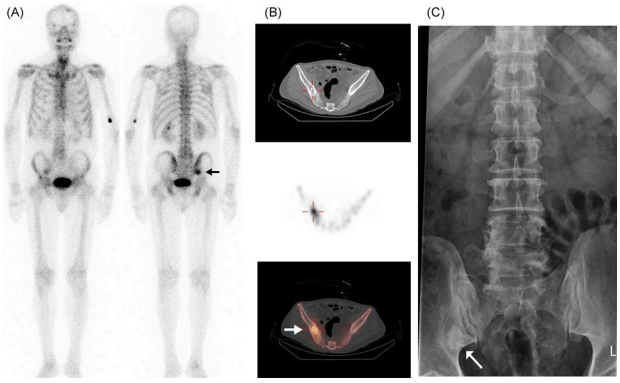

Figure 4. (A) The planar bone scan: a hot spot in lower portion of right ilium. **(B)** The SPECT/CT images: various ossification and cystic formation over right ilium on the reference CT images. **(C)** This finding was not pointed out on the first interpretation of the previous lumbar spine radiography performed 3 years ago from another hospital.

DISCUSSION

Standard bone scanning has long been regarded as a sensitive tool for the investigation of bony metastases. However, the ^{99m}Tc-MDP is not a tumor-specific agent, and primarily, tracer uptake represents the osteoblastic response elicited by a skeletal lesion. Further, this response is nonspecific and can be seen in several benign bony pathologies, such as trauma, infection, and both degenerative and benign bone disease. A variety of conditions may look similar on the bone scan. Careful interpretation is needed for a metastatic work up. Other image modalities or biopsy are usually needed to differentiate malignant metastasis from a benign cause. A number of studies have been published in recent times demonstrating the value of SPECT/CT in this situation. All have shown that SPECT/CT dramatically reduces the number of equivocal/indeterminate bone scans. SPECT/CT increases the diagnostic accuracy of bone scans and significantly decreases the likelihood of a non-diagnostic study requiring further imaging [8-9].

Fibrous dysplasia, in general, appears as an area of markedly increased uptake on the bone scan. The uptake of the radiotracer in the

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affected bones (commonly the craniofacial bones, scapulae, ribs, pelvic bones, spine and extremities) usually occurs in an asymmetric pattern and may be unilateral in the polysostotic variant [5]. CT and MRI are good options to overcome this obstacle easily, by providing accurate anatomical detail. CT is the best technique for depicting lesion extent, cortical boundary and homogeneity of the poorly mineralized lesion. Well defined margination, hazy ground-glass opacity and contrast enhancement are characteristic features of fibrous dysplasia on CT. MRI is quite sensitive for detecting fibrous dysplasia and provides complementary information to CT [9].

These four cases were all asymptomatic and doing fine at the one-year follow-up. Thus no tissue biopsy proof was performed because the following bone scan and radiography revealed no progression of these bone lesions. SPECT/CT increases the diagnostic accuracy of bone scan and the unnecessary surgery or overtreatment of fibrous dysplasia as bone metastasis can be avoided [9,10].

CONCLUSIONS

Although bone scintigraphy has low specificity for fibrous dysplasia, it has a valuable role on identifying disease extent at initial presentation due to its high sensitivity. A whole-body bone scintigraphy at a single session and an additional SPECT/CT, which provides both anatomical and functional data, will be sufficient to elucidate this issue.

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***Conflicts of interest*: Declared none.**

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