

Short Report

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Fortuitously detected Paget's bone disease in a patient with TENIS syndrome on FDG PET/CT

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Case report

A 74-year-old lady presented with a complaint of progressive neck swelling for two years. FNAC from the swelling was suggestive of papillary carcinoma thyroid (PCT). She underwent total thyroidectomy with central compartment lymph node dissection. Histopathology revealed multifocal PCT with central compartment lymph node metastases. Her baseline stimulated sTg was 5.3 ng/ml after levothyroxine withdrawal. WBS showed uptake in the operative bed (not shown). She was treated with 100 mCi of I-131 and was kept on a suppressive dose of thyroxine hormone. Her sTg gradually increased (77.5 ng/ml) in follow-up after three years. WBS was negative for metastatic disease. She underwent FDG PET/CT, that revealed cervical and upper mediastinal lymph nodes. Apart from that, multiple pagetic lesions were also noted. The patient had a history of mild localized pain and tenderness over these. Her Serum Alkaline phosphatase level was elevated (269 U/L). The patient refused resurgery for cervical lymph nodes. She is on Zoledronic acid infusion and remains asymptomatic. She is in regular follow-up, and her suppressed sTg remains stable even after two years.

Thyroid carcinoma is the commonest malignancy of the endocrine system. The majority belongs to well-differentiated thyroid cancers (papillary and follicular cancer) and are relatively

Abstract

Papillary carcinoma thyroid is the prevalent histological subtype of well-differentiated thyroid cancer (WDTC). Patients are treated with thyroidectomy followed by radioactive iodine (RAI) therapy. Follow-up is done with a low-dose whole-body scan (WBS), serum thyroglobulin (sTg), and neck ultrasonography depending upon the stage. Elevated serum sTg despite a negative WBS scan is termed the TENIS (Thyroglobulin elevated negative iodine scintigraphy). FDG PET/CT is indicated in such cases. Here we present an unusual presentation of incidentally detected Paget's disease bone (PDB) in a WDTC presented with TENIS syndrome.

Keywords: Well-differentiated thyroid cancer; TENIS syndrome; Paget's disease bone.

indolent. These have long-term survival in a significant number of nonmetastatic patients. Metastatic thyroid carcinoma is noted in about 10% of patients with papillary carcinoma and up to 25% with follicular carcinoma [1]. The most typical sites for distant metastases are the lungs and bones [2]. The clinical presentation of these patients with differentiated metastatic carcinoma is highly variable. Loss of iodine concentrating capability due to either dedifferentiation or reduction in sodium/iodide symporter expression in postoperative thyroid cancer patients makes iodine inefficient for imaging and therapy [3]. This entity is called TENIS syndrome. In this group of patients, FDG PET/CT has a decisive role. We present a TENIS patient in whom FDG PET/CT helped to find the disease burden and fortuitously detected polyostotic Paget's disease.

The diagnostic and therapeutic approaches for TENIS syndrome do not have a common consensus. It includes observation, re-surgery, chemotherapy, tyrosine kinase inhibitors, re-differentiation therapy, and others [4]. FDG PET/CT is the well-established imaging modality for disease burden evaluation in TENIS patients [5]. Here, FDG PET/CT incidentally detected PDB in a WDTC presented with TENIS syndrome. Paget disease of bone (PDB) is the second most common bone remodeling disease, as osteoporosis leads [6]. PDB is a chronic, pro-

gressive bone disorder that could involve single (monostotic) or multiple bones (polyostotic). The most commonly involved bones include the pelvis, femur, spine, skull, and tibia [7]. Skeletal lesions of PDB are characterized by increased vascularity, osteoclastic bone resorption and, disorganized increased bone formation [6]. Patients with PDB are often asymptomatic. They are frequently incidentally detected with an elevated serum alkaline phosphatase or on X-rays performed for unrelated reasons [8]. Mazières B et al. highlighted the use of scintigraphy in this disease. It allows the spread of the disease to be diagnosed without doing X-rays of the entire skeleton. In twenty percent of cases, this disease cannot be located using X-rays and is detected by scintigraphy. It also supplies an additional objective criterion for judging the development of this disease, especially under treatment [9]. PDB results in weakened bone strength and abnormal bone architecture, leading to pain, deformity or, depending on the bone involved, fracture in the affected bone. The current mainstay of treatment in PDB is bisphosphonates (disodium pamidronate, alendronate, risedronate). They are potent inhibitors of bone resorption and provide prolonged remission of the disease [10]. This case undoubtful utility of the FDG PET/CT in TENIS syndrome. Being a whole-body hybrid molecular imaging, PET/CT helps to unveil many non-malignant lesions.

FDG PET/CT is a valuable imaging modality in the management of the TENIS syndrome. It could unveil several unrelated incidental pathologies such as PDB along with recurrent thyroid cancer.

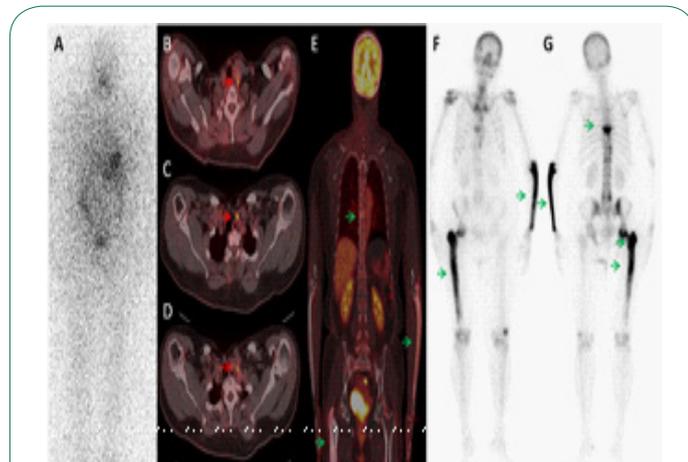


Figure 1: (A) Diagnostic low-dose WBS (anterior) shows physiological tracer distribution. No abnormal tracer avid lesion is noted. (B-D) Transaxial fused FDG-PET/CT shows FDG avid sub-centimeter size left level IV and VI cervical lymph nodes (Red arrow). (F) Whole-body fused coronal FDG PET/CT revealed increased tracer uptake in the body of the D7 vertebra, left radius, and right femur (Green arrow). Significant cortical thickening and sclerosis are noted in these bones. (G) Tc-99m MDP Bone scan (anterior and posterior planar images) revealed intense osteoblastic activity in these bones (green arrow). Scan finding is suggestive of polyostotic PDB.

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