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Clinical vignette

Detection of brown tumors by [^{18}F]F-choline PET/CT performed in a patient with secondary hyperparathyroidism

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Abstract

Brown tumours are rare bone lesions occurring in patients with severe hyperparathyroidism (HPT) because of increased osteoclastic activity due to high levels of parathyroid hormone (PTH). We report the case of 30-year-old woman with secondary hyperparathyroidism due to severe chronic kidney diseases who underwent [^{18}F]F-choline PET/CT scan for localization of the hyperfunctioning parathyroid gland before surgical treatment. [^{18}F]F-choline PET/CT scan showed increased choline uptake in the lower left parathyroid gland and in multiple bone lytic lesions. Multiple focal choline uptake in bone corresponded to brown tumours — fibrous osteitis cystica.

KEY words: [^{18}F]F-choline PET/CT; brown tumours; fibrous osteitis cystica; hyperparathyroidism

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Introduction

Secondary hyperparathyroidism is commonly associated with vitamin D deficiency and chronic kidney disease (CKD). In chronic kidney disease, decreasing GFR leads to decreased phosphate clearance, hyperphosphatemia and hypocalcemia, which then stimulates the parathyroid glands to secrete PTH [1].

Patients with secondary hyperparathyroidism can develop severe complications including renal osteodystrophy, which can further lead to the manifestation of fibrous osteitis cystica — brown tumors. Brown tumors, as unifocal or multifocal bone lesions, form due to a rapid osteoclastic turnover of bone, resulting from the direct effects of PTH.

Despite their name, brown tumors represent a reparative bone process rather than a true neoplasm. Bone loss is replaced by hemorrhage and reparative granulation tissue [2] (Fig. 1). Brown tumors have been reported to occur in 1.5% to 4.5% of patients with primary or secondary hyperparathyroidism [3]. Most commonly it affects young patients, especially in the female population with varying degrees of aggressiveness and risks of recurrence. It can affect the base of the skull, orbits, paranasal sinuses and spinal column as well femur, tibia, humerus, clavicles and scapula. Treatment of a brown tumor aims primarily to reduce the elevated parathyroid hormone by pharmacological treatment or parathyroidectomy. Surgical treatment of brown tumors is reserved for patients with invasive tendencies, painful symptomatology caused by compression on local structures or in patients with high a risk of fracture [4].

At our nuclear medicine department, we are performing [^{18}F]F-choline PET/CT scans regularly for localization of hyperfunctioning parathyroid glands in patients with primary hyperparathyroidism and in consultation with the nephrology department in some cases of severe secondary or tertiary hyperparathyroidism where surgery would be helpful in managing patients' PTH levels.

Case presentation

A 30 years-old female with a case of severe secondary hyperparathyroidism (01./2022 serum Ca 2.39 mmol/L; parathyroid hormone 318 pmol/L) underwent [^{18}F]F-choline PET/CT with a goal of localization of hyperfunctioning parathyroid gland before parathyroidectomy.

In her teenage years, she was diagnosed with SLE (systemic lupus erythematosus) for which she received multiple treatments and was hospitalized on several occasions. In a course of the disease, she developed kidney failure and was put on dialysis. She had

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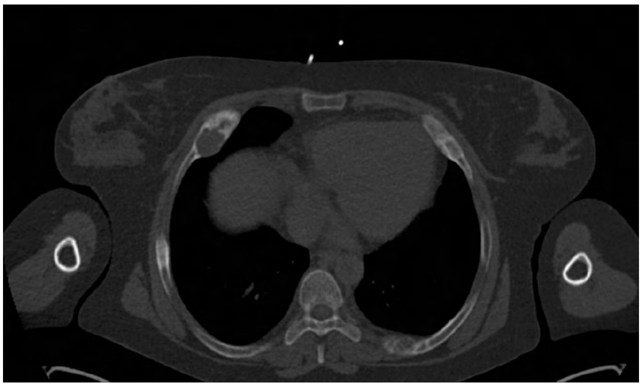

Figure 1. Low dose CT scan showing osteolytic lesions in ribs that are in a fact reparative granulation tissue caused by increased bone turnover

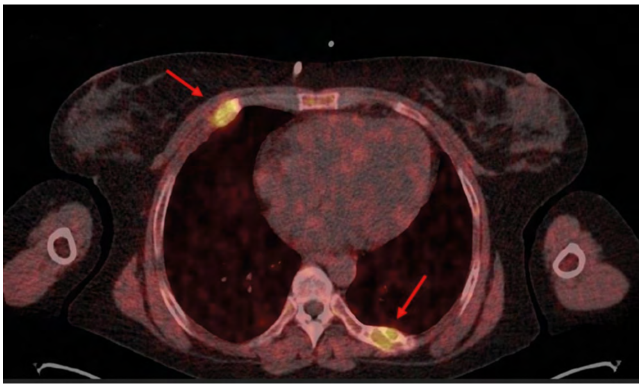

Figure 3. Intense focal choline accumulation in osteolytic rib bone lesions (arrows) (SUV_{max} 8.6 and 10.3) corresponded to brown tumours — fibrous osteitis cystica

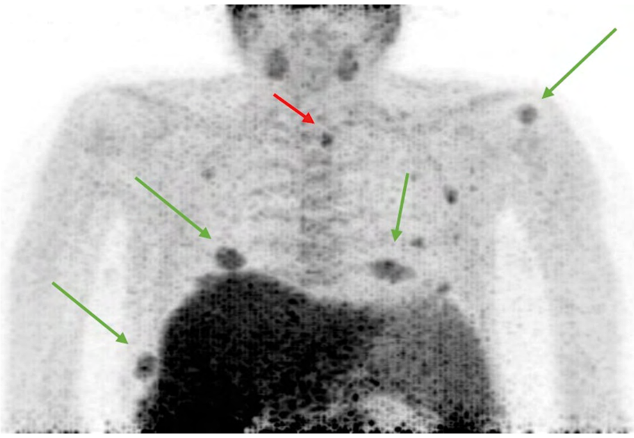

Figure 2. Maximum Intensity Projection (MIP) image from the skull to the diaphragm. Multiple bone lesions with increased focal [^{18}F]F-choline uptake are marked with green arrow. Focal increased uptake in lower left parathyroid gland is marked with red arrow

a kidney transplant but after surgery, she developed a fever and sepsis which caused graft to be removed graft PNF (graft primary nonfunction). Currently, she is receiving dialysis three times a week. On a few occasions, she was hospitalized because of a hypertensive crisis and severe hyperkaliemia. In the last 10 years increased serum levels of PTH were observed, leading to fractures and maxillofacial surgery of tumorous tissue in the upper palate that was described as a brown tissue tumor. With a goal of lowering serum PTH levels and managing complications, the patient was referred to ENT specialist to perform a partial parathyroidectomy.

Subsequently, a [^{18}F]F-choline PET/CT of the neck and mediastinum was performed 10 min after injection of 100 MBq [^{18}F]F-choline PET/CT showed intense focal [^{18}F]F-choline accumulation in a hypodense nodal lesion 1.3 cm in diameter, close to the left lower lobe of the thyroid gland (Fig. 2). Increased focal uptake also was seen in a nodal lesion 0.8 cm in diameter, posterior to the upper right thyroid lobe. The maximum standardized uptake value (SUV_{max}) of the nodal lesion close to the left lower thyroid lobe was 8.2 while 4.3 in the lesion posterior to the right lobe.

Focal uptake was demonstrated in multiple bone lesions, the most evident in the anterior part of right 5th and 9th rib and in the posterior part of left 7th rib. SUV_{max} values were 7.3, 10.3, 8.6 retrospectively (Fig. 2, 3). Intensive accumulation is also seen in the left humeral head (SUV_{max} 5.6).

After positive [^{18}F]F-choline PET/CT patient underwent successful extirpation of the left parathyroid gland with pathohistology confirming it was hyperplastic parathyroid tissue. Serum parathyroid hormone levels were markedly reduced immediately after surgery (preoperative PTH > 318 pmol/L; postoperative 81.95 pmol/L). Serum calcium levels were monitored daily, with calcium carbonate and paricalcitol added to daily therapy. Biochemical markers are still regularly monitored, and the patient continues to receive hemodialysis.

Discussion

Recent studies on the usefulness of [^{18}F]F-choline PET/CT in the localization of hyperfunctioning parathyroid glands in primary hyperparathyroidism have shown that choline scan is highly sensitive and specific in both detection and localization of parathyroid glands (Treglia et al. [5]). There is limited data on the use of choline scans in secondary and tertiary hyperparathyroidism as the treatment approach and management of these patients are different. Parathyroidectomy is only used when conservative methods are exhausted and is reserved for patients with a severe case of hyperparathyroidism.

Brown tumors have shown variable uptake in ^{99m}Tc sestamibi and ^{99m}Tc methylene diphosphonate bone scans (Zanglis et al. [6] and Zhang-Yin J. [7] et al. reported a lack of visualization of brown tumors on ^{99m}Tc-MIBI scintigraphy while on a bone scan Gedik et al. [8] noticed focal and diffusely increased uptake in skeleton). If patients present themselves with a severe case of hyperparathyroidism and have anamnestic data on skeletal lesions or fractures, whole-body PET/CT scan protocol should be used, as skeletal lesions in form of brown tumors can be located anywhere in the axial or peripheral skeleton. This case study demonstrates that [^{18}F]F-choline PET/CT can be a powerful "one-stop shop" for the detection of hyperfunctioning parathyroid glands and brown tumors in patients with secondary and tertiary hyperparathyroidism.

Conflict of interest

The authors declare that they have no conflict of interest.

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