

Case Report

Evaluation of recurrent parathyroid carcinoma: A new imaging tool in uncommon entity

ABSTRACT

Parathyroid carcinoma (PTC) is a rare endocrine tumor and uncommon cause for primary hyperparathyroidism. ^{18}F -fluorocholine (FCH) positron emission tomography and computed tomography (PET/CT) has shown promising results in the detection of parathyroid adenoma, though its role in PTC is undefined due to the paucity of incidence. The authors in this case report discuss the possible complimentary role of FCH PET/CT with ^{18}F -fluorodeoxyglucose PET/CT in the evaluation of the management of suspected recurrence, since this rare entity has high loco-regional and distant recurrence of the disease.

Keywords: ^{18}F -fluorocholine, ^{18}F -fluorodeoxyglucose, positron emission tomography and computed tomography, recurrent parathyroid carcinoma

INTRODUCTION

Parathyroid carcinoma (PTC) – a rare cause of primary hyperparathyroidism requires complete resection of primary disease though it has a tendency of loco-regional/distant recurrence. ^{18}F -fluorodeoxyglucose (FDG) positron emission tomography and computed tomography (PET/CT) has shown its utility in initial staging and restaging of PTC.^[1,2] ^{18}F -fluorocholine (FCH) PET/CT has shown encouraging results in preoperative localization of parathyroid adenoma,^[3,4] but its utility in PTC is limited to a case report.^[5] To the best of our knowledge, this is the second case report, where FCH PET/CT has shown the complimentary role along with FDG PET/CT scan in the evaluation of recurrent disease in a patient with recurrent PTC.

CASE REPORT

A 56-year-old female with elevated serum intact parathyroid hormone (PTH) (3414 pg/ml, normal: 10–65) and calcium levels (12.6 mg/dl, normal: 8.8–10.5) underwent parathyroid surgery a year back for suspicious parathyroid adenoma. The histopathology revealed it as parathyroid cancer (high Ki 67 index, 50%) with vascular and capsular invasion.


The patient declined for redo surgery at that time and presented with left-sided neck swelling, elevated PTH (1027 pg/ml, normal 10–65) and serum calcium levels: (12 mg/dl, normal 8.8–10.5) on follow-up at 1 year later. The serum alkaline phosphatase and creatinine were raised with low serum phosphate levels. The patient underwent technetium-99m-labelled methoxyisobutyl isonitrile (MIBI) dual-phase scintigraphy [Figure 1a and b] and FCH PET/CT [Figure 1c-h] and showed tracer-avid multiple left levels III-IV cervical and the highest mediastinal lymph nodes suggestive of disease recurrence at the loco-regional site.

She subsequently underwent left hemithyroidectomy and modified radical neck dissection. The surgical resected

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Access this article online	
Website: www.wjnm.org	Quick Response Code 
DOI: 10.4103/wjnm.WJNM_67_18	

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How to cite this article: Thanseer N, Parihar AS, Sood A, Bhadada SK, Dahiya D, Singh P, *et al.* Evaluation of recurrent parathyroid carcinoma: A new imaging tool in uncommon entity. World J Nucl Med 2019;18:198-200.

specimen showed PTC infiltrating into adjacent soft tissue and skeletal muscle with multiple lymph nodes metastases on histopathology. She developed breathlessness and pain in the neck after 8 months of the second surgery. She was again evaluated with biochemical parameters and imaging with chest X-ray, FDG, and FCH PET/CT. The chest X-ray showed multiple round soft-tissue densities in bilateral lung fields. The whole-body FDG PET/CT and FCH PET/CT studies [Figure 2a-h] revealed intensely FDG-avid (maximum standardized uptake value (SUVmax) ~13.1) and moderately choline-avid (SUVmax ~ 4.7) multiple parenchymal and pleural-based lung nodules and mediastinal lymph nodes. An additional choline-avid (SUVmax 6.5) and non-FDG-avid lesion in the left parasellar location was also noted, which was suggestive of meningioma on magnetic resonance imaging brain. However, patient again refused for any intervention for the left parasellar lesion. The patient was managed on symptomatic treatment, and she passed

away after being alive for 2 years from the date of her first surgery.

DISCUSSION

The patients of PTC usually present with palpable neck mass, highly elevated PTH, and calcium levels. One-third of the patients may have regional lymph node involvement and metastatic lung, liver, and bone disease and the recurrence of PTC is seen in around 49%–60% of cases.^[1,6] PTC is difficult to diagnose before surgery and usually confirmed on histopathological examination. MIBI scintigraphy cannot differentiate between benign and malignant parathyroid lesions though it has been utilized in localizing the primary and metastatic PTC.^[7] FDG PET/CT has demonstrated to be a valuable imaging modality for the evaluation of PTC regarding tumor metabolism, the extent of the disease and the recurrence at unusual sites, long after primary treatment

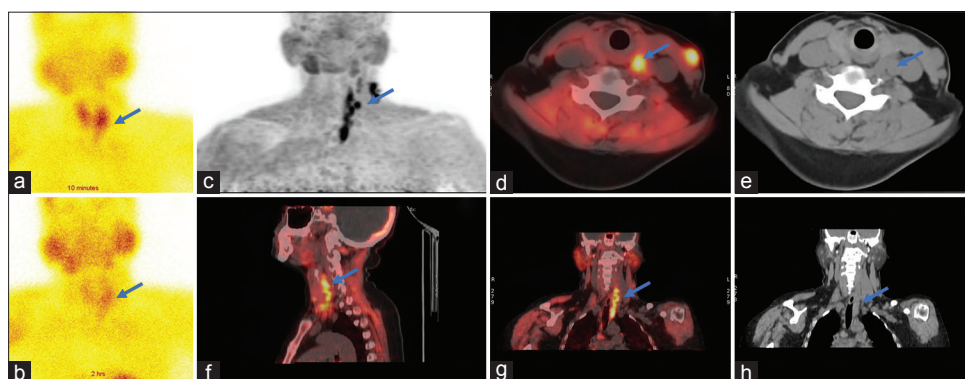


Figure 1: Tc-99m methoxyisobutyl isonitrile dual phase scintigraphy at 10 min (early) and 2 h (delayed) showing tracer-avid lesion in the left neck (a and b). ¹⁸F-fluorocholine positron emission tomography and computed tomography maximal intensity projection, cross-sectional computed tomography and fused images for recurrent disease evaluation, showing tracer-avid multiple left cervical lymph nodes (c-h)

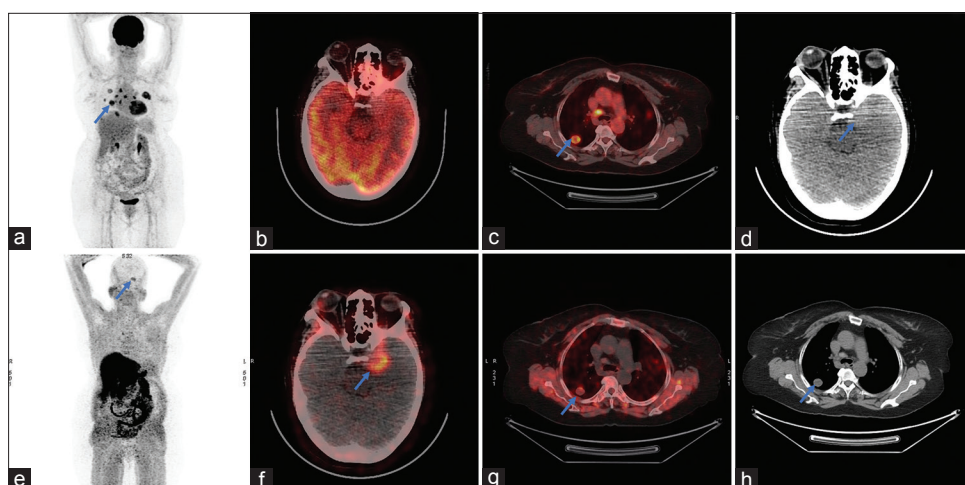


Figure 2: ¹⁸F-fluorodeoxyglucose positron emission tomography and computed tomography and ¹⁸F-fluorocholine positron emission tomography and computed tomography maximal intensity projection, cross-sectional computed tomography and fused images showing intensely ¹⁸F-fluorodeoxyglucose-avid (standardized uptake value maximum ~13.1) and moderately choline-avid (standardized uptake value maximum ~4.7) multiple parenchymal and pleural-based lung nodules and mediastinal lymph nodes (arrows in figures a, c, e, g and h). A choline-avid (standardized uptake value maximum 6.5) and non-¹⁸F-fluorodeoxyglucose-avid lesion in the left parasellar location (arrows in figures b, d and f) MIP: Maximal intensity projection

where conventional imaging failed in picking the disease. The primary disease and metastatic sites have shown significant FDG avidity. The increased SUV value generally indicates the aggressive behavior of the tumor and poor prognosis.^[2,8,9] In the index case, the preoperative MIBI showed tracer-avid left inferior parathyroid lesion followed by postoperative imaging done with MIBI and FCH studies at 1 year showed intense tracer uptake in the recurrent primary lesion, cervical and mediastinal nodes. However, the FDG and FCH imaging performed after a period of 8 months following second surgery showed high FDG and low choline uptake in the mediastinal lymph nodes and lung nodules. The reason for the divergent avidity of two tracers probably could be that the recurrent disease had de-differentiated by that time. The choline positive and FDG negative left parasellar lesion was suggestive of meningioma. Similarly, in a study of 300 patients of prostate cancer submitted for FCH PET/CT scanning for staging or restaging, six patients showed additional focal FCH uptake in meningiomas.^[10]

FCH PET/CT has shown promising role in the preoperative localization of parathyroid adenoma,^[3,4] but its role in PTC is limited to a case report where additional metastatic lesions were detected both by FCH and FDG PET/CT in treated patient of PTC emphasizing the complementary role of two imaging techniques. The authors postulated that difference in tumor differentiation and proliferation might have led to discordance in FCH and FDG uptake in metastatic lesions.^[5] Our case had shown the intense cervical lymph nodal FCH uptake at first recurrence, but moderate FCH and intense FDG avidity in metastatic pulmonary and mediastinal nodal disease in the subsequent recurrence with the possibility of tumor de-differentiation during that interval. The FCH PET/CT may have a complementary role in addition to FDG PET/CT in staging or restaging of PTC in demonstrating the location and extent of the recurrent disease for further management. The role of FCH PET/CT needs to be explored in PTC work-up.

FCH PFCH PET/CT as an imaging modality may have an adjunctive role in patients presenting with parathyroid cancer in staging and restaging since this rare entity has high loco-regional and distant recurrence of the disease.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understand that names and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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