Trails on ^{18}F-Fluorodeoxyglucose Positron Emission Tomography/Computed Tomography Leading to Diagnosis of Testicular Adrenal Rest Tumor

Abstract

Testicular adrenal rest tumors (TARTs) are secondary to hypertrophy of adrenal rest cells in the rete testis in settings of hypersecretion of androgens. We present a case of congenital adrenal hyperplasia with TART with clues to the diagnosis on ^{18}F-fluorodeoxyglucose positron emission tomography/computed tomography (^{18}F-FDG PET/CT). To the best of our knowledge, this is the first reported case on the role of ^{18}F-FDG PET/CT in TART.

Keywords: *^{18}F-fluorodeoxyglucose, positron emission tomography/computed tomography, testicular adrenal rest tumor*

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Introduction

Interpretation of 18F-FDG PET has pitfalls from areas of benign physiological and pathological uptake of glucose. However, the same areas of physiological uptake can be secondary changes that, many a time, yield clues to underlying disease process. We present one such case where the FDG distribution yielded clue to disease process which was not known prior to the PET scan and thereby significantly altering the course of the management of the patient.

Case Report

A 17-year-old male was diagnosed with bilateral testicular mass lesions. The right orchidectomy was done, and histology was interpreted as Leydig cell tumor. The patient was referred for restaging with an ^{18}F-fluorodeoxyglucose positron emission tomography/computed tomography (^{18}F-FDG PET/CT). The whole-body image shows increased uptake in the left scrotum [Figure 1a]. Incidentally, there was unusual and prominent increased uptake in the adrenal glands on both sides [Figure 1a]. The corresponding CT images showed a significant hypertrophy of the adrenal glands [Figure 1b] with increased metabolism [Figure 1c]. The lesion in the left testis showed homogeneous enhancement measuring 1.5 cm × 2.6 cm with irregular margin and standardized uptake value (SUVmax) of 4 [Figure 2a and b]. Review of clinical history revealed short stature height of 147 cm and early onset of pubic hair at 8 years age. The patient also demonstrated orthostatic hypotension with systolic blood pressure in the recumbent position being 130 mmHg, and on standing, it was 100 mmHg. The 17-hydroxyprogesterone levels were elevated on subsequent evaluation (>20 ng/ml). The presence of bilateral testicular lesions, with adrenal hypertrophy, features of androgen excess in the form of early pubarche, short stature, and biochemical evidence of raised 17-OH progesterone leads conclusively to diagnosis of congenital adrenal hyperplasia (CAH) with testicular adrenal rest tumor (TART). The patient was referred to an endocrinologist for semen analysis and further management.

Discussion

TARTs are benign tumors. They are associated with CAH. Up to 94% of patients with CAH can have TARTs.[1] They are typically present in rete testis and commonly bilateral. Usually, CAH is due to congenital defect in the 21-hydroxylase enzyme which results in aldosterone deficiency. This activates the feedback and resulting in increased pituitary secretion of Adrenocorticotropic hormone that results in adrenal hypertrophy and overproduction of adrenal androgens.

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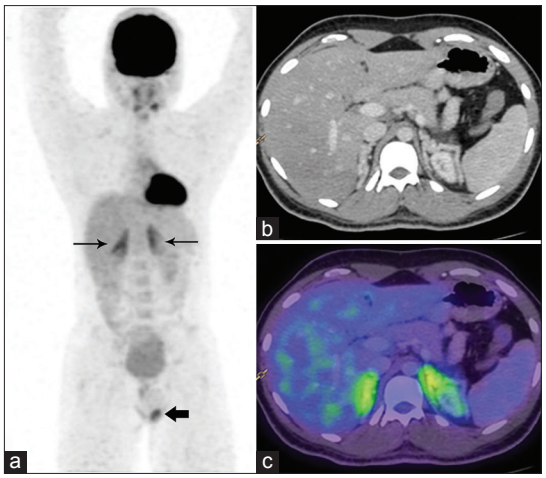

Figure 1: (a) Whole-body fluorodeoxyglucose image showing the left testicular lesion (bold arrow) and bilateral adrenal hypermetabolism, (b) computed tomography showing grossly hypertrophied adrenal glands (c) showing fused positron emission tomography/computed tomography image demonstrating adrenal hypermetabolism

When large, these tumors result in obstructive azoospermia and infertility. TARTs are often misdiagnosed as Leydig cell tumors due to similar features on pathology resulting in a diagnostic dilemma.[2] The presence of tumors bilaterally and biochemical features can aid in appropriate diagnosis. Leydig cell tumor tends to show relatively higher metabolic activity[3] while the left testicular lesion in our study showed mildly increased uptake compared to the liver (SUVmax 4).

In patients with TARTs, glucocorticoid suppression can result in regression of the growth. The patient should be evaluated for infertility, and orchidectomy is reserved until irreversible changes are noticed in the testis secondary to obstruction from the tumor. Testis-sparing surgery is also considered in these patients.[4] Rich *et al*.[5] advocate an "aggressive conservative approach" in children diagnosed with Leydig cell tumors and that orchidectomy should not be attempted without complete endocrine profile in these patients.

In our case, the presence of abnormal hypertrophied adrenal glands in the whole-body image showing

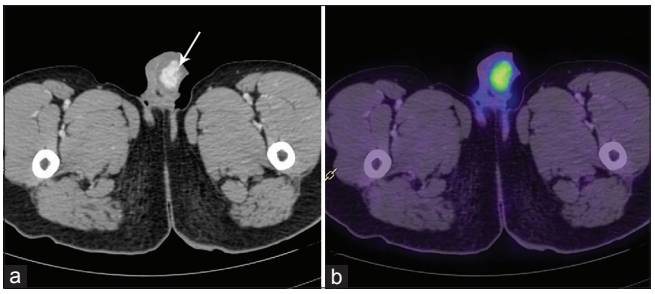

Figure 2: Computed tomography (a) and fused positron emission tomography/computed tomography (b) images of the left testicular lesion showing homogeneously enhancing lesion with increased metabolic activity

hypermetabolism was the clue that leads to re-examination of the entire clinical history and changing the diagnosis of the patient and changing the treatment plan. To the best of our knowledge, this is the first reported case on the role of ^{18}F-FDG PET/CT in TART.

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Conflicts of interest

There are no conflicts of interest.

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