

CASE REPORT

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Leydig cell tumor of the testis: an incidental finding at 18F-FDG PET/CT imaging

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Abstract

Background: Leydig cell tumors (LCTs) represent the most common form of stromal tumors. We reported the 18F-fluorodeoxyglucose (FDG) positron emission tomography (PET)/computed tomography (CT) findings of a patient with testicular LCT.

Case presentation: A 50-year-old man with a history of end-stage renal disease and renal transplantation 19 years ago. One year earlier, he started to have a chronic rejection. During the investigation to determine the cause of chronic rejection, a suspicious lesion in the graft with a collection around it was seen on ultrasound (US) images, raising the possibility of post-transplant lymphoproliferative disorder (PTLD). The patient was referred for further evaluation by whole body 18F-FDG PET/CT imaging. The image finding revealed an incidental hypermetabolic focal lesion in the right testicle—no other specific findings in the remaining parts of the body nor definitive FDG avid lymphadenopathy to suggest PTLD. Testicular US was requested and showed a well-defined right-sided heterogeneous hypoechoic intratesticular focal mass at the upper pole of the right testis with significant internal vascularity on the color Doppler imaging. The patient underwent a right radical orchidectomy, and the tumor was pathologically confirmed as an LCT.

Conclusion: In our case, 18F-FDG-PET/CT has been helpful in incidentally detecting this rare testicular tumor in a patient with suspected PTLD.

Keywords: Leydig cell tumors, 18F-FDG PET/CT, Renal transplantation

Background

Although LCTs are rare, it is considered the most frequent non-germ cell tumors of the testis, accounting for 1–3% of all adult testicular tumors and 0.4–9% of prepubescent testicular tumors [1, 2]. LCTs are usually unilateral. In most cases, they originate from testicular Leydig cells, but rarely they may arise from the epididymis [3]. A small percentage of LCTs are malignant and have a poor metastatic prognosis since they react poorly to chemotherapy and radiation treatment [1]. Adults may have unusual symptoms associated with testicular tumors and endocrine problems or may remain entirely

asymptomatic and be detected accidentally by ultrasonography (US) [4].

The US has shown high sensitivity in identifying testicular abnormalities among all imaging modalities and is considered the gold standard technique for evaluating the testes. However, relying only on gray-scale and color Doppler information is inadequate to describe lesions, particularly small lesions, in which LCTs and seminomas may be quite similar [5, 6]. LCTs usually exhibit a homogenous hypoechoic appearance in the testicular US imaging. Heterogeneity may be indicative of aggressive behavior [7].

The use of 18F-FDG PET/CT may not assist in distinguishing between benign and malignant testicular tumors, but it is helpful in managing and monitoring an LCT patient. FDG PET/CT has a very high negative predictive value in FDG-avid testicular tumors, but its

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Fig. 1 a–c FDG PET/CT images demonstrate incidental finding of focal increase right testicular uptake seen with more metabolic activity in relation to the remaining testicular tissues (SUV_{max} 2.9). No specific findings in other areas or definitive FDG avid lymphadenopathy

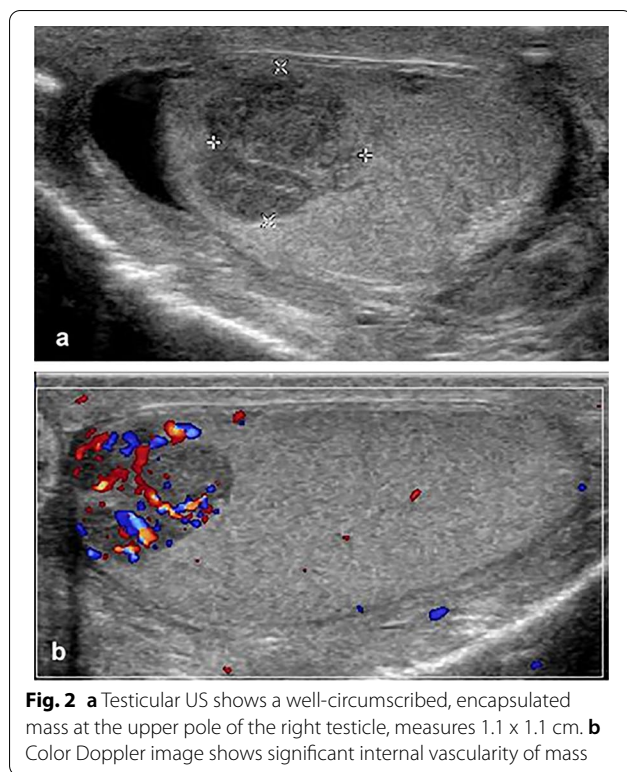


Fig. 2 a Testicular US shows a well-circumscribed, encapsulated mass at the upper pole of the right testicle, measures 1.1 x 1.1 cm. b Color Doppler image shows significant internal vascularity of mass

positive predictive value varies across studies [8]. FDG PET/CT findings of LCTs have been reported in few studies [9, 10]. The present clinical case report described the 18F-FDG PET/CT findings of testicular LCT detected during imaging of a patient with the possibility of post-transplant lymphoproliferative disorder (PTLD).



Fig. 3 An orchidectomy specimen was received with an attached spermatic cord, revealing a 1 x 1 x 1 cm, well-defined mass on serial cut sections. The surface is tan homogenous in an unremarkable testicular background

Case presentation

A 50-year-old man with a history of end-stage renal disease and renal transplantation 19 years ago. One year earlier, he went into chronic rejection and started on hemodialysis. During the investigation by the nephrology team to determine the cause of chronic rejection, a suspicious lesion in the graft with a surrounding collection was clear on the US examination, raising the possibility of PTLD. The nephrology team referred the patient for further evaluation by 18F-FDG PET/CT examination. The 18F-DG PET/CT was done and revealed an incidental finding of focal increase right testicular uptake with more metabolic activity ($SUV_{max} = 2.9$) in relation to the remaining testicular tissues (Fig. 1). No specific findings in other areas nor definitive FDG avid lymphadenopathy to suggest PTLD. The testicular US was requested and showed a corresponding focal mass at the upper pole of the right testicle (Fig. 2a) with significant internal vascularity on color Doppler imaging (Fig. 2b). During that time, the patient showed no testicular swelling, testicular pain, weight loss, or a decrease in appetite. The patient was referred to the urology department and underwent a right radical orchidectomy. A right inguinal incision was made, the spermatic cord was exposed and dissected, which was normal looking, and then ligation of the vas deferens and vessels was done at the internal inguinal ring. The right testis was exposed and looked normal in size and contour compared to the left one. The right epididymis looked unremarkable. The testis was then dissected and delivered with the cord included in the tunica. Homeostasis was maintained. After the operation, the patient did well with no complaints. He is currently under active surveillance.

An orchidectomy specimen with the attached spermatic cord was sent for histopathology. The specimen revealed a 1 x 1 x 1 cm well-defined mass on serial cut sections. The surface was homogenous tan (Fig. 3). The tumor displayed a diffuse pattern of large polygonal cells with round nuclei, profuse eosinophilic cytoplasm, and prominent nucleoli (Fig. 4a). No definite Reinke crystals were identified. There was no evidence of tumor necrosis, nuclear pleomorphism, increased mitosis, or vascular invasion. The Ki67 proliferative index was low, with 1–2% (Fig. 4b). The tumor cells were diffusely positive for vimentin, inhibin, and calretinin and focally positive for Mart-1 (Figs. 4c–f). The spermatic cord showed benign adrenal rest and Leydig cell hyperplasia. The findings were consistent with benign LCT.

Discussion

PET/CT has been widely used to identify and discriminate benign and malignant tumors in various organs [11, 12]; however, a few papers have been published on

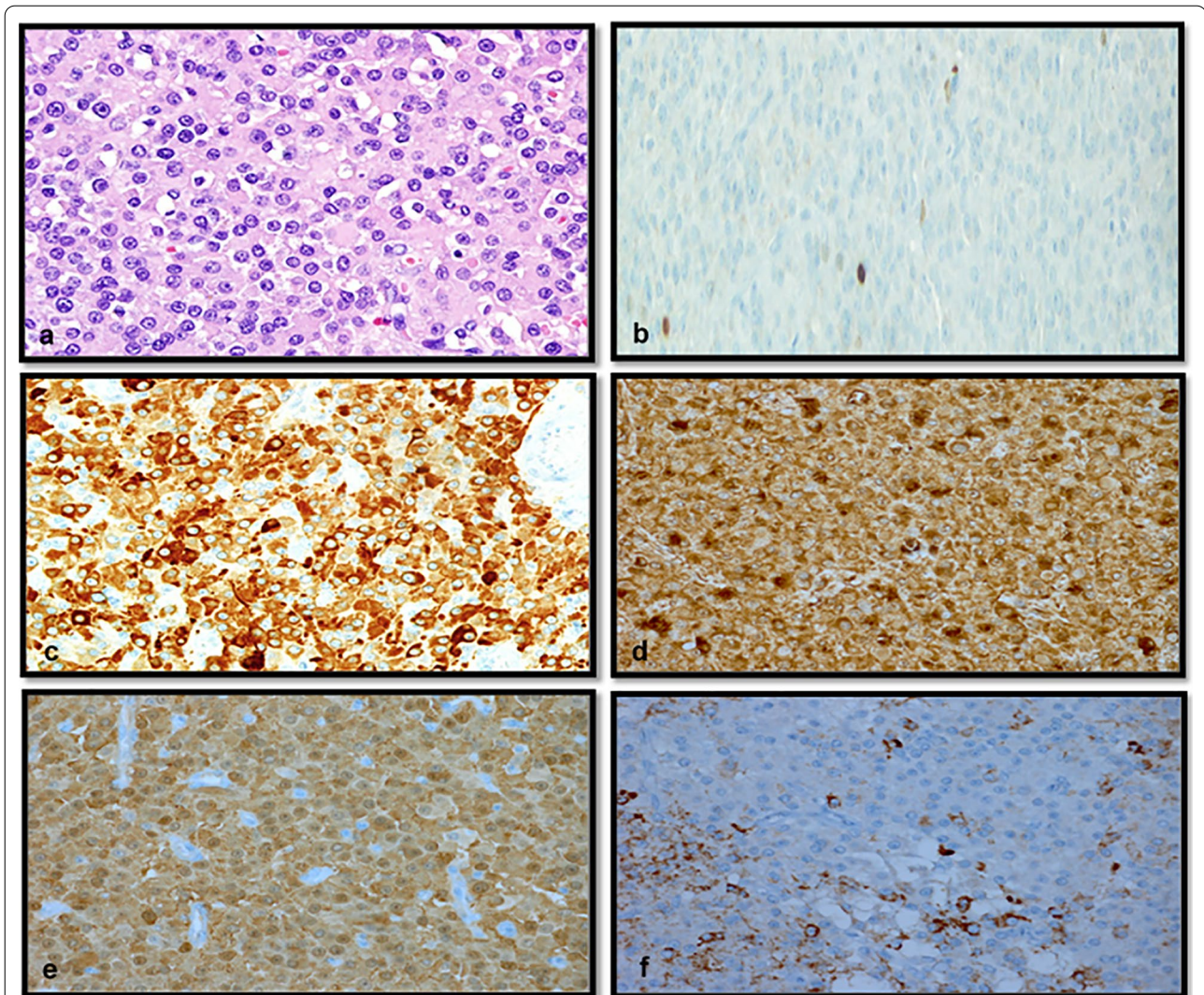


Fig. 4 **a** Light microscopic examination of the tumor shows a diffuse pattern of large polygonal cells with abundant eosinophilic cytoplasm, round nuclei, and prominent nucleoli. No definite Reinke crystals were identified. There is no evidence of tumor necrosis, nuclear pleomorphism, increase mitosis, or vascular invasion. **b** The Ki67 proliferative index is low with 1–2%. **c** The tumor cells are diffusely positive for inhibin. **d** The tumor cells are diffusely positive for vimentin. **e** The tumor cells are diffusely positive for calretinin. **f** The tumor cells are focally positive for Mart-1. The spermatic cord shows benign adrenal rest and Leydig cell hyperplasia. The diagnosis was consistent with benign LCT

its application to detect testicular lesions and assess the functional status of testes [13]. Our case illustrated the ^{18}F -FDG PET/CT findings of an LCT in a 50-year-old male patient who had a renal transplant 19 years ago. The ^{18}F -FDG PET/CT demonstrated an incidental finding of focal increase right testicular uptake with more metabolic activity in relation to the remaining testicular tissues ($\text{SUV}_{\text{max}} = 2.9$).

A few studies have addressed the PET/CT imaging of LCT using ^{18}F -FDG. One of these case studies was a 43-year-old man submitted to whole-body ^{18}F -FDG PET/CT imaging for healthcare screening. The ^{18}F -FDG

PET/CT imaging showed a focal hypermetabolic lesion in the right testis. The tumor was pathologically verified as LCT [9]. Another case of a 50-year-old man with lymphoma was described, in which increased ^{18}F -FDG uptake was noted in a right testicular lesion. After right radical orchidectomy, a benign LCT was histopathologically proved [10].

SUV_{max} is a semi-quantitative biomarker with a particular clinical reference value for benign and malignant tumor distinction [14, 15]. Many factors affecting testicular SUV such as age, testicular volume, and sexual activity [16]. Discrepancies in normal SUVs may exist because of

differences in patient demographics and PET technology [17]. A study included 203 men showed that the normal SUV ranges between 1.23 and 3.85 with a mean value of 2.44 [18]. Shoe et al. [19] discovered statistically significant variations in SUVmax values for benign and malignant testicular tumors. SUVmax of 3.75 was found to be the optimal cutoff value for distinguishing malignant from benign testicular lesions; this value has a sensitivity of 90.6% and a specificity of 80.9%. In our study, the right testis showed more focal uptake than the left one (SUVmax = 2.9). The left testis was seen with unified, homogeneous uptake, and the SUVmax was in the normal range (SUVmax = 2.0).

In our case, testicular uptake of FDG prompted a medical referral to a urologist consultation and scrotal US. The US was done and demonstrate a well-defined heterogeneous hypoechoic focal mass at the upper pole of the right testicle with significant internal vascularity on color Doppler imaging, which corresponding to the focal hypermetabolic lesion in the right testicle on 18F-FDG PET/CT imaging. However, neither intratumoral calcification nor testicular microlithiasis was observed in US images. A review of one article describing the US findings of testicular LCTs shows that 90% of the cases are homogeneous hypoechoic, and 88% have a distinctive pattern of increased peripheral blood flow [20].

Conclusion

In our case, 18F-FDG-PET/CT was valuable in incidentally detecting this uncommon testicular tumor in a patient with suspected PTLD. Further researches are required to determine the mechanism of FDG uptake and the role of FDG PET/CT in this tumor type.

Abbreviations

CT: Computed tomography; FDG: Fluorodeoxyglucose; LCTs: Leydig cell tumors; PET: Positron emission tomography; PTLD: Posttransplant lymphoproliferative disorder; SUV: Standardized uptake value; US: Ultrasound.

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Author's contributions

Author read and approved the final manuscript.

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Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethical approval and consent to participate

Institutional review board's approval was obtained. Written informed consent was obtained from the patient.

Consent for publication

Not applicable

Informed consent

Written informed consent was obtained.

Competing interests

The authors of this case report declare no relevant conflicts of interest, and no relationships with any companies, whose products or services may be related to the subject matter of the article.

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