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Case Report
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Evaluation of Yolk sac tumor in a pediatric patient with F-18 FDG PET/CT

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ABSTRACT

Yolk sac tumors are a form of malignant primitive germ cell tumor that exhibit similarities to the mesenchymal cells of the primitive yolk sac in histological terms. These tumors are frequently observed in pediatric patients and can be found in various locations throughout the body. It is essential to diagnose and treat this condition promptly to minimize the high morbidity and mortality associated with it. The aim of this report is to assess the efficacy of F-18 FDG PET/CT in the initial staging of the disease, evaluating chemotherapy response, and restaging the disease following chemotherapy and a second surgical intervention.

Keywords: Yolk sac tumor, pediatric patient, PET/CT

INTRODUCTION

Malignant germ cell tumor (MGCT), of which yolk sac tumor (YST) is the most common subtype, accounting for only 1-4% of childhood malignancies.[1] YST is thought to arise from primitive germ cells. YST can be of testicular, ovarian or extragonadal origin.[2] The level of alpha-fetoprotein (AFP) is high in most patients due to the endodermal origin of YST. Therefore, the level of AFP is considered to be an important biomarker.

The outcome of patients with YST is closely related to the stage of the disease. Studies have shown that children with gonadal YST in stage 1 may benefit from surgical intervention. F-18 FDG PET/CT is a system that integrates metabolic imaging with anatomical imaging. F-18 FDG PET/CT is widely used in pediatric lymphoma, neuroblastoma, soft tissue sarcoma and other malignancies. However, the use of F-18 FDG PET/CT in pediatric YST has mostly been reported in case reports.[3] In this report, we evaluated the usefulness of F-18 FDG PET/CT in the initial staging of the disease, in the assessment of chemotherapy response, and in the restaging of the disease after chemotherapy and second surgical treatment.

CASE REPORT

A 15-year-old Syrian girl patient underwent surgery for abdominal swelling and pain in her country. A right salpingo-oophorectomy was performed. A mass measuring 220x150x120 mm was removed. Pathology report compatible with an ovarian YST.

Then the patient was admitted to our hospital for treatment. Blood tests showed AFP 426 ng/ml (reference range 0-40 ng/ml), Beta HCG 0.1 mIU/ml (reference range 0-10 mIU/ml). Abdominal and pelvic MRI, F-18 FDG PET/CT imaging and USG were performed. A total of four F-18 FDG PET/CT scans of a pediatric patient were analysed.

F-18 FDG PET/CT images were evaluated retrospectively by two nuclear medicine physicians by consensus. Both were blinded to the clinical and imaging findings. F-18 FDG PET/CT images were interpreted as positive or negative for disease. Any non-physiological focal F-18 FDG uptake on PET was anatomically localised on CT. Semi-quantitative analysis was performed using the maximum standardized uptake value (SUV max).

On evaluation of the first post-operative F-18 FDG PET/CT scan, multiple hypermetabolic lesions were observed on the left side of the pelvis on the MIP image. Cross-sectional imaging at the level of the pelvic inlet showed metastatic foci in the right rectus abdominis muscle and a metastatic focus near the left psoas muscle were hypermetabolic. Pelvic masses anterior and posterior to the uterus and a mass in the anterior wall of the left pelvis were hypermetabolic and pelvic fluid was observed (**Figure 1a, 1b**). At the level of the pelvic floor, increased F-18

FDG uptake was seen in a hypodense lesion in the left rectus abdominis muscle and in inguinal lymph nodes.

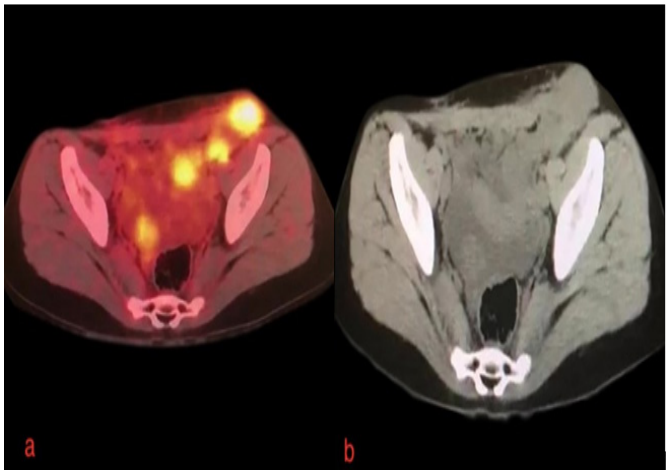

Figure 1. First post-operative F-18 FDG PET/CT images. Fusion image (**a**) and CT image (**b**)

The cisplatin-etoposide-bleomycin (PEB) chemotherapy protocol was used. The second follow-up F-18 FDG PET/CT scan was performed after 2 cycles of the PEB protocol. In the last F-18 FDG PET/CT scan, no pathological FDG uptake was observed in the pelvic region on MIP images. Cross-sectional imaging at the level of the pelvic inlet showed a calcified lesion without significant FDG uptake in the right rectus abdominis muscle. No mass was observed in the left lateral psoas muscle. At the level of the uterus, there was no significant FDG uptake in the calcified lesions of the left anterior pelvic wall, and no other lesions of the pelvis were observed (**Figure 2a, 2b**). **Table** shows the temporal change in the number, size and metabolic activity of the lesions after initial surgery, following response to chemotherapy and at the end of treatment by F-18 FDG PET/CT imaging and the decrease in serum AFP levels.

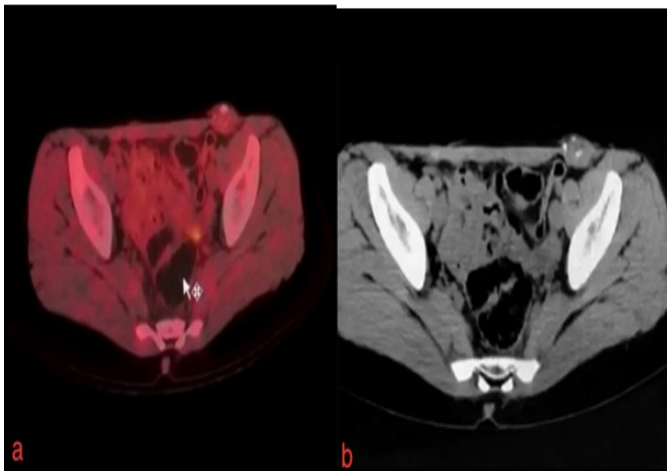

Figure 2. F-18 FDG PET/CT images at the end of chemotherapy. Fusion image (**a**), CT image (**b**)

Following MRI findings of a slight increase in the size of a calcified mass in the anterior wall of the pelvis, a decision was made at a consultative meeting to excise the mass and surgery was performed. The patient's current pathology was compatible with a mature teratoma (**Figure 3**). A decrease in AFP level is shown in **Figure 4** and **Table**.

Mature teratomas are considered benign and are not included in the management protocol. The patient is currently stable and under close observation. No residual disease was found on the second post-surgery imagings. No distant metastases were observed.

Table. The temporal characteristics of the lesions (size, number, location, SUVmax) and serum AFP levels

Study no	LTS (mm)/ no of lesion	Lesion sites	SUVmax	AFP (ng/ml)
1	39x23 mm/5 33x19 mm/6 33x3 mm/4	Pelvic anterior wall Pelvic Inguinal	4.2 4.6 6.8	426
2	30x17 mm/5 16x16 mm/1 Ss/multiple	Pelvic anterior wall Pelvic Left external iliac	1.0 2.8 1.8	21.15
3	27x15 mm/5 12 mm/1 Ss/multiple	Pelvic anterior wall Pelvic Left external iliac	2.4 1.9 3.8	1.91
4	26x16 mm/5 - -	Pelvic anterior wall Pelvic Left external iliac, left inguinal	- - -	1.77

AFP: Alpha-fetoprotein, LTS: Largest tumor size, Ss: Subsantimetric

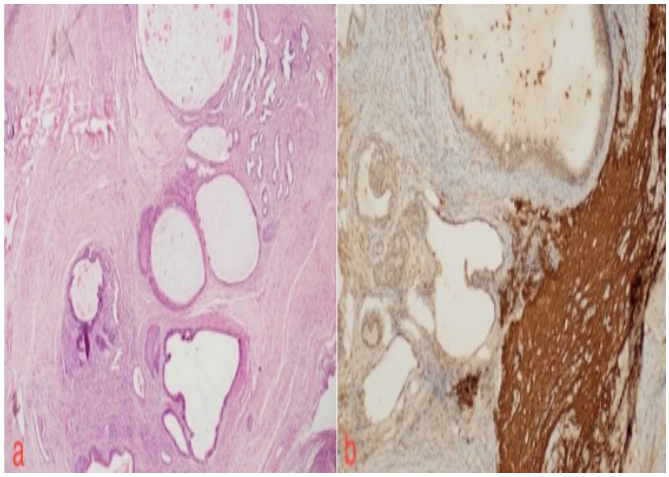

Figure 3. Histopathology compatible with a mature teratoma. Hematoxylin and eosin staining (HEx10) (**a**). Glial tissue stained positive for GFAP, immunohistochemical staining (IHKx10) (**b**)

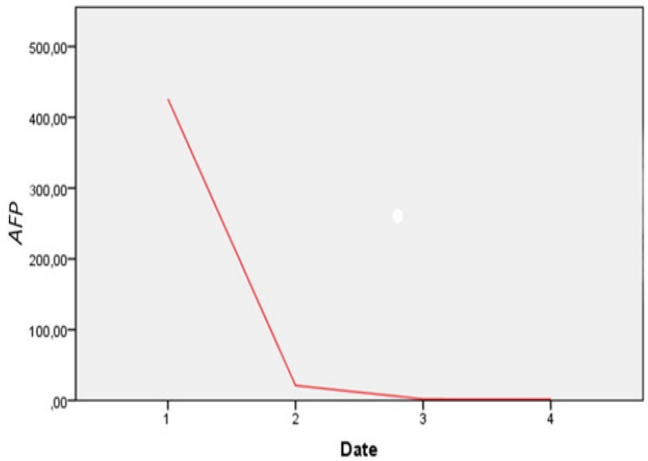

Figure 4. Follow-up AFP measurement levels
AFP: Alpha-fetoprotein

DISCUSSION

YST is a rare tumor that is often included in the MGCT subgroup. Few studies in the literature have focused on YST alone.[4] Most studies in the literature have focused on MGCT, a heterogeneous group that includes seminoma, dysgerminoma,

embryonal carcinoma, choriocarcinoma, polyembryoma, gonadoblastoma and teratoma.[5]

There are few studies on the usefulness of F-18 FDG PET/CT in MGCT, and most of these have been in the adult population.[6] There are even fewer studies on the use of F-18 FDG PET/CT in pediatric MGCT. It is important to clarify the usefulness of F-18 FDG PET/CT in pediatric YST.[7] The value of F-18 FDG PET/CT in preoperative staging of pediatric YST or MGCT is unknown. F-18 FDG PET/CT is not routinely recommended for initial staging of MGCT in adults because of the small additional benefit of F-18 FDG PET/CT compared with CT or MRI.[8] In our study, although F-18 FDG PET/CT provided additional metabolic information to conventional imaging, the F-18 FDG PET/CT findings did not change the stage of the disease and therefore did not change clinical management in the initial studies. However, last F-18 FDG PET/CT images contributed to the decision to refer the ametabolic pelvic anterior wall mass for surgery. Histopathological examination showed that the lesion was compatible with benign mature teratoma.

CONCLUSION

F-18 FDG PET/CT is an imaging modality with high diagnostic accuracy in demonstrating anatomical and metabolic response to chemotherapy after surgical treatment in patients YST. It can show tumor differentiation at follow-up and has a major impact on therapeutic management.

ETHICAL DECLARATIONS

Informed Consent

The patient's parents signed a free and informed consent form.

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

The authors declared that this study has received no financial support.

Author Contributions

All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

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