Case Report

Granulosa cell tumor induced massive recurrence of post hysterectomy leiomyoma

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ABSTRACT

The authors report a very unusual occurrence of a massive recurrence of leiomyoma from post hysterectomy stump diagnosed on fluorine-18 fluorodeoxyglucose positron emission tomography/computed tomography (F-18-FDG PET/CT). The case also has an additional complexity of granulosa cell tumor (GCT) of ovary probably contributing to the recurrence and massive size.

Keywords: Estrogen, F-18- fluoro deoxy glucose, granulosae cell tumor, lieomyoma

INTRODUCTION

Uterine leiomyomas reaching enormous size is not an unusual occurrence. Recurrence of leiomyomas occurs infrequently due to variable causes. Attributable causes rarely include granulosa cell tumor (GCT) of ovary, due to probable induced estrogen stimulation. Post hysterectomy recurrence of leiomyoma from the cervical stump and the incidental association of GCT make a very rare association in the causation of massive recurrence diagnosed on fluorine-18 fluorodeoxyglucose positron emission tomography/computed tomography (F18-FDG PET/CT).

CASE REPORT

An asymptomatic, 44-year-old, primiparous woman on routine examination for medical fitness was found to have a large lower abdominal mass. The mass was nontender, not ballotable, nor freely mobile, and there was no free fluid in the abdomen. Her last child birth was 18 years back by lower segment cesarean section (LSCS). She underwent total abdominal hysterectomy 16 years back for massive uterine fibroids extending high up into the upper abdomen with a histopathological confirmation of benign leiomyoma. Ultrasonography performed reported as hysterectomy status with a large 17 × 11 cm hypoechoic pelvic mass of left ovarian origin and right ovary appearing enlarged measuring 5.7 × 3.4 cm with multiple, thin-walled cysts with a maximum size of 2.8 × 2.2 cm. No free fluid in the pelvis or abdomen. T2-weighted magnetic resonance imaging (MRI) pelvis revealed post hysterectomy status and a hypointense lobulated mass 15 × 13 × 10 cm in the left side of pelvis extending up to fourth lumbar vertebral level with a 3 × 2 × 2 cm cystic mass adherent to the main mass [Figure 1]. Carcinoembryonic antigen (CEA) was elevated with 20.2 pg/ml and alpha fetoprotein (AFP), cancer antigen (CA) 125, and CA-15.3 were with in normal limits. In view of the large pelvic mass and elevated CEA, a F-18-FDG PET/CT of abdomen was performed. Transaxial, sagittal, and coronal reformatted images revealed a non-FDG, avid, uniform-density, large mass with lobular contour arising from pelvis isodense to muscle and showing continuity with the anterior cervical wall. No abnormal calcifications or necrosis was noted within the mass. The mass was abutting the left posterolateral vesicle wall pushing the bladder to the right and superiorly. The fat planes with adjoining rectum and vesicle wall were well-maintained [Figure 2a and b]. Visualized ovary appeared enlarged measuring 6.0 × 4.5 cm with multiple cystic areas within and adherent to the abdominopelvic mass. No FDG avidity was seen in the ovarian mass [Figure 3]. In view of the homogeneous and myomatous texture of the mass being strikingly non-FDG avid and the mass being traceable and contiguous with the cervical stump, possibility of a metabolically inactive benign pathology of recurrent leiomyoma was considered despite a hysterectomy status. Patient underwent laparotomy which showed a large pelvic mass with multiple lobulations and

Access this article online

Quick Response Code:

Website: www.ijnm.in

DOI: 10.4103/0972-3919.136584

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adherent to the bladder, viscera, and the anterior abdominal wall. The mass could be easily dissected from the adjoining structures and excised completely along with the ovary adherent to the mass posteriorly. Postoperative period was uneventful and patient discharged on the 4th postoperative day. Gross specimen showing a large, homogeneous, mural mass with a septated cystic ovarian mass was seen adherent posteriorly. Histopathology of the mural mass revealed intersecting short fascicles of smooth muscle cells with intervening abundant collagen and no mitosis or necrosis, features suggesting benign Leiomyoma [Figure 4a and b]. The attached ovarian lesion revealed a 4 cm mass with fleshy cut sections and the tumor composed of cohesive sheets of cells showing focal trabecular pattern. These cells had vesicular oval nuclei, longitudinal nuclear grooving, and minimal eosinophilic cytoplasm. Increased mitosis or necrosis was not seen. The tumor was concluded as granulosa cell tumor-adult type [Figure 5a and b]. In view of the metabolically bland lesion comprising of normal uterine muscularity and the associated cystic ovarian mass being low-grade, well-differentiated, GCT; no further treatment was envisaged and the patient is on follow-up with no evidence of any disease.

DISCUSSION

Uterine leiomyomas are ubiquitous in occurrence with enigmatic enormity of size and variable age of occurrence being fairly well-encountered.[1,2] Leiomyomas arise from the myometrium of the uterine intramural area from a single clone of smooth muscle cells, with continued growth in one direction. Uterine fibroids are estrogen dependent and may reach different sizes and more frequently could be multiple.[3] Total abdominal hysterectomy or myomectomy are the commonly practiced mode of treatment depending on the size and extent of the disease. Recurrence is often noted with infrequency. Documented etiology of uterine leiomyomas is estrogen and progesterone, ovarian steroids, insulin growth factor-1 (IGF-1), and prolactin (PRL). However, in almost all cases the attributable cause being idiopathic and the exact cause unfathomed. Estrogen seems to be the active incriminating factor involved in increasing the size of fibroids.[4,5] Presence of coexisting GCT of the ovary in this reported case substantiates the functional dependence of fibroid growth on estrogen and explains probable reason for the recurrence from a minute residual smooth muscle of the post total hysterectomy stump and the mammoth size attained. GCTs are sex cord stromal group of tumors made up of granulosa cells, theca cells, and fibroblasts in varying degrees and combinations.[5] GCTs account for approximately 2% of all ovarian tumors and can be divided into adult (95%) and juvenile (5%) types based on histological findings. Both subtypes commonly produce

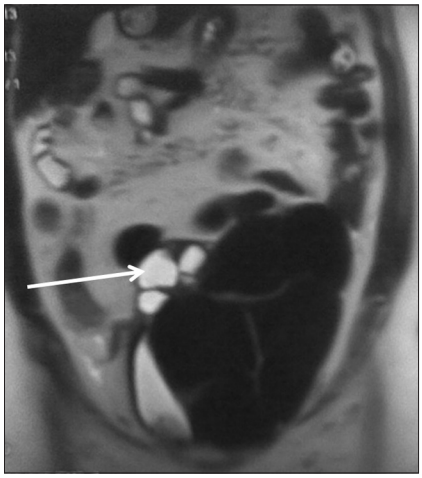

Figure 1: Coronal T2-weighted (T2W) magnetic resonance imaging (MRI) pelvis showing hypointense lobulated solid mass displacing the bladder laterally to the right with a cystic mass adherent to it (arrow)

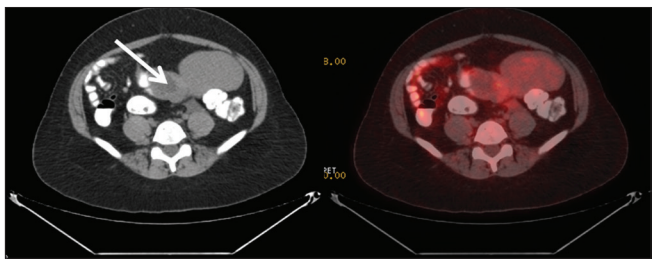

Figure 3: Axial PET/CT images of pelvis showing non-FDG enlarged ovary with cystic areas adherent to the pelvic mass (arrow)

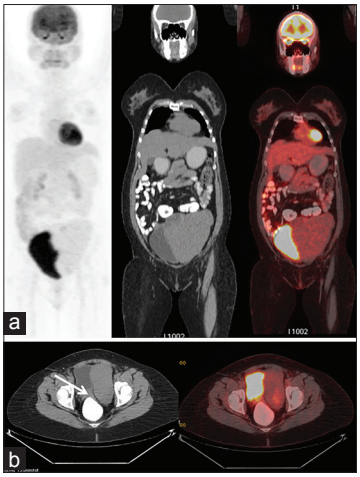

Figure 2: (a) Maximum intensity projection (MIP) and coronal positron emission tomography/computed tomography (PET/CT) images of pelvis showing a large, non-fluorodeoxyglucose (FDG), avid, uniform-density, solid mass isodense to muscle. (b) Axial sections showing continuity of the mass with anterior cervical wall (arrow)

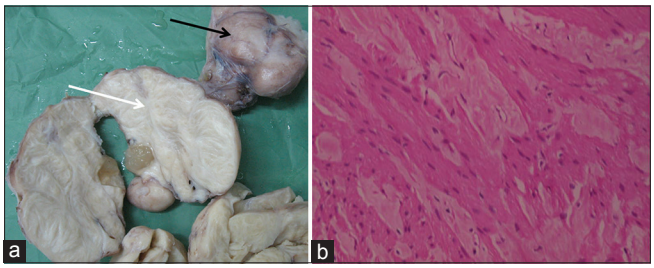

Figure 4: (a) Gross specimen showing a large homogeneous mural mass (arrow) with a cystic ovarian mass seen adherent posteriorly (black arrow). (b) High power views of the mural mass showing intersecting short fascicles of smooth muscle cells with intervening collagen suggesting leiomyoma

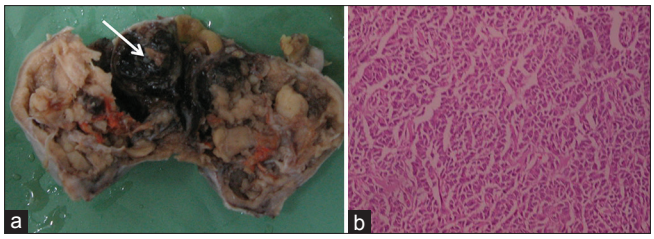

Figure 5: (a) The attached ovarian lesion showing fleshy cut sections with septated cystic and hemorrhagic areas (arrow). (b) High power view showing cohesive sheets of cells with focal trabecular pattern having vesicular oval nuclei, longitudinal nuclear grooving, and minimal eosinophilic cytoplasm, suggesting granulosa cell tumor-adult type

estrogen. However, while adult GCT (AGCT) usually occur in postmenopausal women and have late recurrences, most juvenile granulosa cell tumor (JGCT) develop in individuals younger than 30 years.[6] The cellular typing of the GCT in our case being a low-risk category, places the patient in good prognosis.[6,7] With complete removal of the recurrent fibroid along with attached ovarian tumor in totality, the chances of further recurrence of fibroids are almost nonexistent. The patient is disease free on follow-up.

CONCLUSION

The case highlights the unusual recurrence of uterine fibroid from the cervical stump in a post total hysterectomy status and the recurrent fibroid being diagnosed based on the PET/CT features of nonmetabolic nature and the CT features revealing a normal solid myomatous texture of the lesion traceable to the cervical stump.[8] Other non-FDG, avid lesions like mucinous cystadenomas show septate and cystic component which is conspicuous by its absence in this case. Association of incidentally detected GCT from the ovary attached to the recurrent myoma strengthens the etiopathological attribute of the recurrence of fibroid to the GCT. Hormones secreted by GCT were the probable cause of recurrence and the enormity of size attained.

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How to cite this article: Chalanki MV, Dattatreya S, Padmaja P, Dayal M, Parakh M, Rao VS. Granulosa cell tumor induced massive recurrence of post hysterectomy leiomyoma. Indian J Nucl Med 2014;29:179-81.

Source of Support: Nil. **Conflict of Interest:** None declared.