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Case Report

A rare case of extragonadal immature teratoma mimicking as subserosal pedunculated myoma

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ABSTRACT

Immature teratomas are usually derived from a malignant transformation of mature teratoma. The pure immature teratoma accounts for less than 1% of all ovarian cancers. It is the second most common germ cell malignancy and accounts for 10-20% of all ovarian malignancies seen in women younger than 20 years of age. Extragonadal origin are extremely rare and the most common extragonadal site of these teratomas is the omentum. We hereby describe a case report of a 29-year-old lady who presented with abdominal pain and her imaging with an ultrasound revealed a mass with features suggestive of a subserosal fibroid. She underwent a laproscopic myomectomy. A histopathologic diagnosis of Immature teratoma was made following her primary surgery. She subsequently underwent a staging laparotomy which was followed by chemotherapy. Immature teratomas predominantly occur in young patients, and preservation of fertility is an important factor in its management. Treatment should be initiated as soon as possible after surgery, preferably within 7-10 days, in those patients who require chemotherapy.

Keywords: Alpha-fetoprotein, Dermoid cyst, Extragonadal, Greater omentum, Immature teratoma, Subserousal myoma

INTRODUCTION

Germ cell tumors are congenital tumors frequently seen in gonads, containing derivatives of all the three germinal layers (ectoderm, mesoderm, endoderm). Teratoma is the most commonly encountered germ cell tumor among the ovarian tumors. Mature cystic teratomas (benign cystic teratoma or dermoid cysts) are among the most common ovarian tumors. The immature form was first described in 1960 by Thürlbeck and Scully, and can be pure or mixed with a mature component.[1]

Immature teratomas are usually derived from a malignant transformation of mature teratomas.[2] The amount of neuroectodermal immature tissue present permits the classification of immature teratomas into three grades of increasing malignancy. The pure immature teratoma accounts for less than 1% of all ovarian cancer, but it is second most common germ cell malignancy and accounts for 10-20% of all ovarian malignancies seen in women younger than 20 years of age.[3] The occurrence of teratoma in males is less common than in females. Immature ovarian teratoma may be associated with gliomatosis peritonei, which has a favourable prognosis if composed of completely mature tissue. Immature teratomas of extragonadal origin are extremely rare. The most common extragonadal site of these teratomas is the omentum.

CASE REPORT

A 29-year-old woman who was para one live one (P1L1) was admitted to our hospital with complaint of lower abdominal pain since 2 months. Pain was on and off, dull aching in type, not related to activity and was not relieved after taking analgesics. Her menstrual cycles were regular with normal blood flow. Her past medical history was not significant for any medical comorbidities. On admission,

bimanual examination was suggestive of normal uterine size. There was no tenderness in either adenexal region. Per speculum examination revealed a normal cervix and vagina with no discharge. Ultrasound of her pelvis showed a bulky uterus (10×4.1×6.6 cm) with a large pedunculated heterogenous mass with foci of calcification noted arising from the fundus of uterus (Figure 1). Features on imaging were suggestive of a subserosal pedunculated fundal uterine fibroid (6×4.3×4.8 cm). Bilateral ovaries were normal. Preoperative laboratory test results were within normal limits. After a pre-operative evaluation, a decision was taken for laproscopic myomectomy.

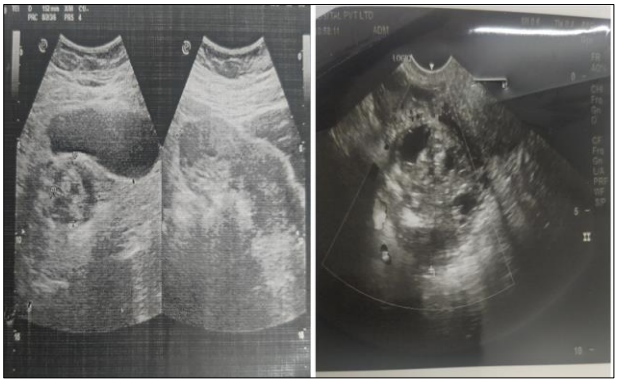

Figure 1: Ultrasound of a heterogenous mass with foci of calcification.

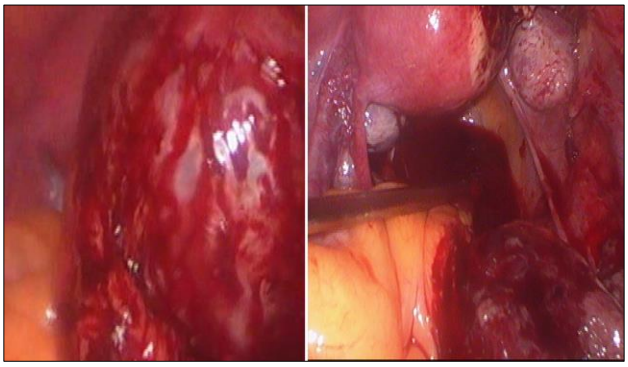

Figure 2: Intraoperative finding of a mass adherent to greater omentum. The ovaries on both sides appeared normal.

Intraoperative findings

On laproscopy, a single, solid cystic mass of size 7×7 cm was found, adherent to the bowel and omentum posteriorly and to the fundus of the uterus anteriorly. Right side of the fallopian tube was stretched out on the mass. Bilateral ovaries looked grossly normal and the pouch of doughlas (POD) was obliterated with mass. The mass was encapsulated, smooth with glistening outer surface (Figure 2).

It was excised partially from the omentum and completely from the uterus. Pathological examination of the excised tumour confirmed a typical pattern of immature teratoma grade III, comprising of skin with adipose tissue, bony fragments, intestinal epithelium, retinal elements, mature and immature neural tissue elements which occupied more than three low power fields (Figure 3). Post-procedure tumour markers were done which revealed elevation of alfa fetoprotein (26.67 ng/ml). The other tumour markers were normal (CA125, CA19-9, carcinoembryonic antigen). Magnetic resonance imaging (MRI) abdomen and pelvis, computed tomography (CT) chest and CT brain were done to rule out metastasis.

Hence a decision was taken for staging laprotomy after which the patient proceeded for total abdominal hysterectomy (TAH) with bilateral salpingo-oopherectomy with infra colic omentectomy and retroperitoneal lymph node dissection (RPLND). Patient tolerated the procedure well and made a good post-operative recovery. The histopathology was suggestive of normal ovaries with no lymph nodal involvement. In view of both ovaries being normal and not showing any evidence of tumour microscopically, two possibilities can be drawn to ascertain its origin- 1) Extra gonadal tumour 2) Accessory ovary. Patient was discharged on post-operative day five in good general condition and was referred to gynec-oncologist for chemotherapy.

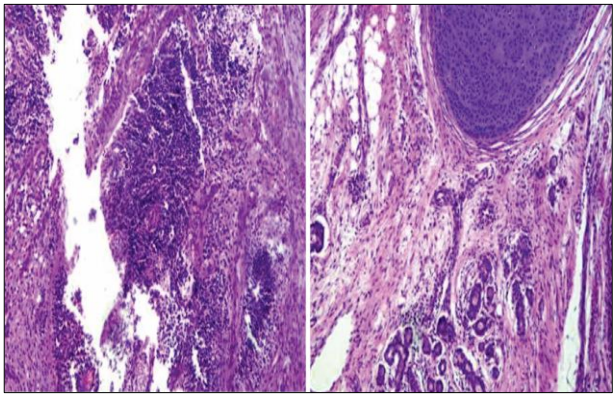

Figure 3: Histopathology suggestive of immature teratoma.

DISCUSSION

Immature teratoma contains elements that resemble tissue derived from embryo. The pure immature teratoma accounts for less than 1% of all ovarian cancers, but it is the second most common germ cell malignancy. It accounts for 10-20% of all ovarian malignancies seen in women younger than 20 years of age.[3] About 50% of pure immature teratomas of the ovary occurs in females between the age of ten and twenty years and rarely occurs in postmenopausal women.

Teratomas arise from germ cells that fail to mature normally in the gonadal locations. These totipotent cells can differentiate into tissue components representing

derivatives of mesoderm, ectoderm and endoderm. The distribution of teratomas are described in order of decreasing frequency: in the ovaries, the testes, the anterior mediastinum, the retroperitoneal space, the presacral and coccygeal areas, pineal and other intracranial sites, the neck and abdominal viscera other than the gonads.[4] The migratory property of germ cells would explain teratomas in these extragonadal sites, which generally occur along midline structures. Extra gonadal immature teratoma is a very rare condition. Teratomas of the greater omentum have been reported more frequently in women. They are typically found in women of reproductive age and may also appear in young girls and older women.

Teratomas arise from germ cells that generally originate in the mature gonads. During early fetal development, germ cells migrate from yolk sac along the hindgut toward the genital ridge i.e. primitive gonad. These totipotent cells may give rise to a variety of tissues originating from the three primitive embryonic layers. Migration along the hindgut explains how teratomas may develop in multiple locations. The etiology of extra gonadal teratomas is poorly understood, but 3 main theories have been proposed to explain their location:

- Primary teratomas of the omentum originate from displaced germ cells.
- Teratomas may develop in a supernumerary ovary of the omentum.
- Teratomas may result from autoamputation of an ovarian dermoid cyst with secondary implantation into the greater omentum.

It is difficult to establish a diagnosis preoperatively. In our case, the patient was initially diagnosed with a subersosal pedunculated degenerated calcified fibroid based on her imaging and was later diagnosed to have grade III immature teratoma based on her histopathological report. Metastatic workup (CT chest, MRI pelvis and CT brain) was negative in our patient and thus a decision was taken for chemotherapy.

Smith et al, reported a case of a 68-year-old woman who presented with a mass and uterine prolapse.[5] Radiological investigations revealed two intraabdominal cystic lesions with calcification. Surgical exploration, resection and histopathology suggested a coexisting omental and ovarian teratoma with ovarian neoplasm. Khoo et al, reported a similar case of a 29-year-old Chinese woman who presented with lower abdominal pain, vaginal discharge and fever.[6] She was clinically diagnosed to have pelvic inflammatory disease. Ultrasound later suggested two cystic lesions.

Laparoscopic ovarian cystectomy intraoperatively detected another omental mass in the POD, which was resected. The histopathology of both showed mature teratoma. Kearney et al, reported a case of synchronous benign teratoma of the omentum and ovary in 1983.[7] The case report is of a 70-year-old woman who died of massive myocardial infarction. Postmortem examination revealed a left ovarian dermoid cyst and a coexisting greater omental teratoma which was histologically proved later.

Kubosawa et al, reported a case of a 62-year-old woman who had a laparotomy and bilateral salpingooophorectomy, found to have multiple cystic teratomas of the omentum.[8] The histological picture showed invasive adenocarcinoma within the lining of the cyst and also with peritoneal dissemination. To conclude, immature teratomas predominantly occur in the young patients, and the preservation of fertility is an important factor in its management. The most important feature of the immature teratoma is the grade of the lesion. In addition, the stage of disease and extent of tumor at the initiation of treatment also has an impact on its prognosis. In patients with stage 1A and grade I tumours, it is sufficient to perform a staging surgery with a unilateral oophorectomy. Patients with a higher grade (II or III) tumour with stage 1A immature teratoma or a more advanced stage disease should be treated with adjuvant chemotherapy containing bleomycin, etoposide and cisplatin in addition to surgery.

Treatment should be initiated as soon as possible after surgery, preferably within seven to ten 10 days in those who require chemotherapy. Overall, the five year survival rate has been reported to be 82%, 62%, 30% for patient with grade 1, 2 and 3 respectively.[3] Thus a timely diagnosis and prompt treatment dictates the prognosis in such patients.

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