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Research Paper

Ovarian dysgerminoma with torsion: case report and review of the literature

A. QABA, H.OUADIA,F. INTIDAME, K. HAROU, A. SOUMMANI[1]
1 Department of Obstetrics and Gynecology, Mohamed VI University Hospital

Abstract
We report a case of a 16-year-old girl diagnosed with an ovarian dysgerminoma accompanied with adnexal torsion who presented with abdominal mass, pain but no recurrent fever. After a series of supportive treatment, we performed the surgery at the best time. Then the patient received 3 cycles of chemotherapy. The patient followed up regularly after chemotherapy. Until the article is submitted, the patient is still alive, no significant sign of recurrence was found in her regular follow-up.

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I. Introduction:

An ovarian dysgerminoma is a rare, malignant tumor occurring in young women, accounting for 1% to 2% of all primary ovarian neoplasms [1]. Although rare, dysgerminomas are the most common malignant ovarian germ cell tumor, accounting for approximately 33% to 38% of all malignant ovarian germ cell tumors.

Dysgerminomas originate from the primordial germ cells of the embryonic gonads and due to their origin, are histologically equivalent to the testicular seminoma [1]. Clinically it presents as abdominal mass with or without abdominal pain.

Early surgery is important for patients with dysgerminoma, especially for those accompanied with ovarian torsion.

Because a majority of patients with dysgerminoma are young women, preservation of fertility should always be considered.

For stage 1A dysgerminomas, a fertility-preserving surgery without adjuvant chemotherapy or radiotherapy is usually enough as studies have shown.

II. Case Report:

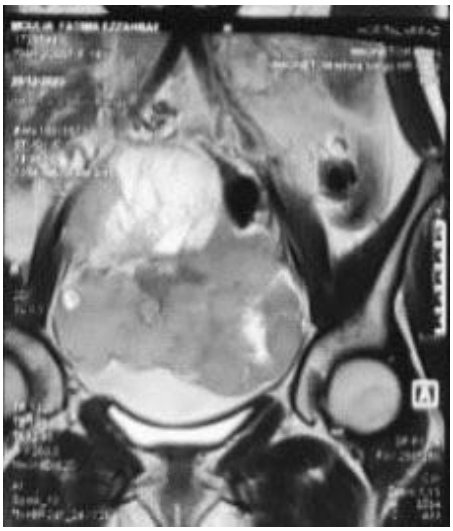
A 16-year-old female, presented to our emergency room with lower abdominal pain for 10 days. The pain was not accompanied with any other sigh such as fever, nausea or constipation.

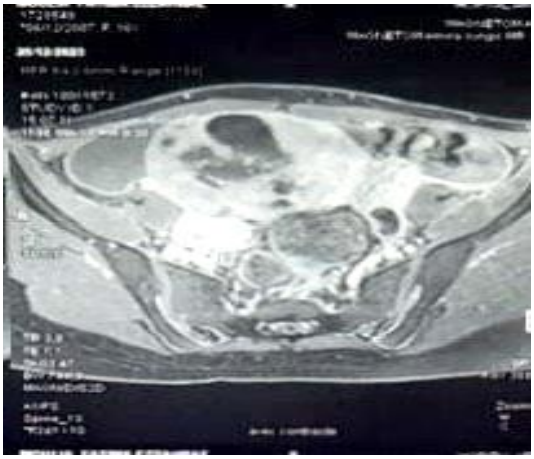
Menstrual cycles were regular with a last menstrual period of three weeks prior. Physical examination found a palpable firm mass located at the hypogastric region of the abdomen.

Quantitative β-HCG was 234 mIU/mL (normal, <3 mIU/mL), Alphafetoprotein (AFP) was normal. Transabdominal sonography showed a normal-sized uterus. No gestational sac was identified within the endometrium. A large, heterogeneous, solid centropelvic mass was detected measuring 11*11cm. the mass was thought to have arisen from the right adnexal region. The left ovary was normal in size and appearance. A significant amount of pelvic fluid was present.

3 D T1 sequence before and after dynamic gadolinium injection T1 VIBE revealed a large ovarian centro-pelvic mass measuring 11 x 7.5 x 11 cm (TxAP x CC) with lobulated contours, heterogeneous tissue signal, T1 isosignal, intermediate signal intermediary T2 diffusion hypersignal with low ADC, moderately contrast-enhanced delineating large areas of necrosis.

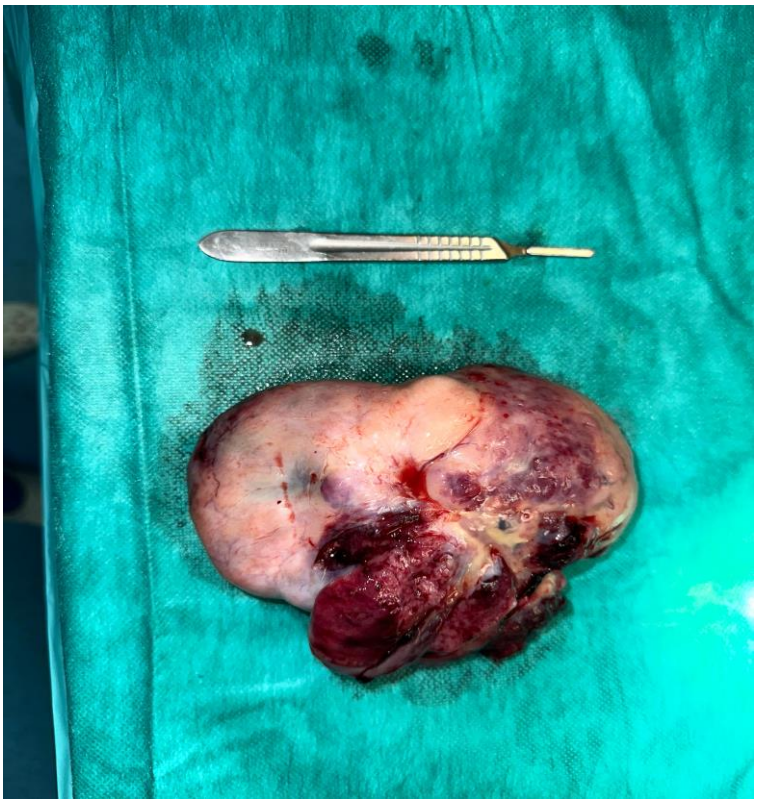
Between the uterus and the lesion process described above, a vascular mass with multiple serpiginous structures forming a spiral of two turns opposite the ovarian hilum.





Description: Axial cine MRI showing a large heterogeneous mass lesion in the right adnexa with twisting of the ovarian vascular pedicle. Origin: Department of Radiology, MED VI hospital MARRAKECH

Based on the patient's age, laboratory values, and radiographic findings, the most likely diagnosis was ovarian dysgerminoma. One day later, the patient underwent exploratory laparoscopic surgery, a right salpingo-oophorectomy was conducted. The pathologic diagnosis was returned as dysgerminoma of the right ovary, Stage IA.
Follicular preservation was proposed before starting the chemotherapy protocol but the patient and her parents refused due to lack of financial means.



macroscopic appearance of the adnexectomy

III. Discussion:

Although dysgerminoma accounts for only 2% of all ovarian tumors, it accounts for 33% of malignant ovarian germ cell tumors [2,3]. In addition, three-fourths of dysgerminomas arise in young adults and adolescents [2,4], especially women under age 30, however, they can be found in all ages, even found during pregnancy [4,5].

Dysgerminomas originate from the primordial germ cells of the embryonic gonads and due to their origin are histologically equivalent to the testicular seminoma [6,7].

The most common clinical presentation of an ovarian dysgerminoma is a young patient with subacute, lower quadrant pain and a palpable pelvic mass by clinical examination. Symptom duration is short and associated with rapid tumor growth. Acute pain related to torsion or rupture is uncommon [6,7,8]. When ovarian torsion occurs, edema and inflammation can lead to the expansion of the ovary. It has been reported that adnexal torsion of ovary is usually benign, malignancy rate in ovarian torsion was about 2%, and most of them were in stage I [9,10].

The most common and convenient used preoperative imaging diagnostic method is ultrasound [11]. After searching extensive literature, it indicated that the typical sonographic appearances of ovarian dysgerminoma were huge, solid, adnexal mass with lobulated and irregular echogenicity within it, accompanied with abundant blood flow signals [11]. Moreover, magnetic resonance imaging (MRI) and CT may also provide auxiliary diagnosis basis.

Tumor markers may aid in the diagnosis and postoperative monitoring of ovarian dysgerminomas. Tumor marker elevation is dependent on the type of tissue components that comprise a mass. Elevation ofβ-HCG and serum LDH can be seen in association with a dysgerminoma. Elevated LDH is noted in most cases of ovarian dysgerminomas. Alpha-fetoprotein values, which are elevated in some types of malignant ovarian germ cell tumors, are negative with dysgerminomas. Quantification of the tumor markers β-HCG and LDH at diagnosis allows for monitoring of tumor recurrence after treatment [8,12].

Despite the fact that all dysgerminomas are malignant, they do have excellent prognosis after a simple salpingooophorectomy up to 96% cure rate of a unilateral tumor without capsular invasion or spread. And because of its excellent response to chemotherapy, those that have extended beyond the ovary can often be cured, with overall survival of greater than 80%.

In treating dysgerminoma, surgery is not only therapeutic but also required for diagnosis and staging, with scope of procedure dependent on intraoperative findings and patient's desire—whether or not she wants to maintain fertility or avoid exogenous estrogen. Patient management is focused on treating the tumor while preserving fertility in these patients. Tumor staging based on the International Federation of Obstetrics and Gynecology (FIGO) finds 65% to 75% of patients are classified with Stage IA dysgerminoma with disease limited to one ovary and with no metastatic involvement at the time of diagnosis [15,16].

The standard treatment for Stage IA dysgerminoma is a fertility-preserving surgical approach: unilateral salpingo-oophorectomy with staging biopsies and preservation of the uterus and contralateral ovary. The literature varies on whether a biopsy should be done of a normal appearing contralateral ovary. It may detect occult disease, but it may also affect future fertility due to adhesions or ovarian failure [17].

Patients with completely resected stage IA disease usually receive 3 cycles of bleomycin, etoposide, and cisplatin while higher stages receive 4 cycles [13,14]

Five-year survival rate for Stage 1A dysgerminoma is greater than 90% to 95%. Stage 1A disease treated surgically does require a close clinical follow-up and monitoring of relevant laboratory values. The Recurrence of Stage 1A dysgerminomas is rare (10%-15%), typically occurs during the first two years after diagnosis, and responds well to chemotherapy [6,9,12].

Of the several classes of chemotherapeutic agents, those most related to ovarian toxicity are alkylating agents. Furthermore, when combination therapy is applied, the effect of any single agent is more difficult to measure [18,19].

The accurate mechanism by which a given chemotherapeutic agent provokes ovarian injury is inadequately understood. Chemotherapy has been linked to depletion of primordial follicles.

During the treatments, ovaries are subjected to DNA damage and apoptosis leading to accelerated follicles loss and infertility.

The influence of chemotherapy on follicular health in term of the first cell types affected by chemotherapy are varied among studies [20]. Cisplatin has been proven to trigger the apoptosis events in granulosa cells within primordial follicles, resulting in increased follicle loss. The magnitude of follicular loss is interconnected with the accumulative dose and duration of chemotherapy [18].

IV. Conclusion:

Although dysgerminomas with adnexal torsion is extremely rare, the diagnosis should be taken in consideration when a patient presents with solid adnexal mass and acute abdominal pain. Ultrasound, CT, MRI and tumor markers may provide some auxiliary diagnosis basis.

Early surgery is necessary for dysgerminomas and the fertility-preserving surgical approach with unilateral salpingo-oophorectomy represent the standard treatment for Stage IA.

Patients with dysgerminoma usually have long overall survival.

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