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Case Report

Bilateral ovarian mature teratoma with unilateral torsion in a 42 years old female in a low-income country

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ABSTRACT

Bilateral ovarian mature teratomas are rare. They occur mostly in the reproductive age group women. Serious complications such torsion required prompt surgical intervention. No case so far has been reported in the Haitian literature. We present a case of a 42-years-old female arrived at the Centre Medical Frere after a two-day delay due to limited healthcare access, experiencing acute abdominal pain and vomiting. After clinical and ultrasound examination, we concluded to the diagnosis of ovarian torsion with patient having bilateral ovarian mass which guided us considering different parameters to a total hysterectomy with salpingo-oophorectomy. Histopathology confirmed bilateral mature teratomas with torsion. This case helps bring awareness regarding the importance of regular gynecologic follow-up, along early detection and management especially in resource-limited settings where delayed care can lead to complications.

Keywords: Bilateral ovarian teratoma, Delayed healthcare, Histopathology, Ovarian torsion, Surgical emergency

INTRODUCTION

Mature ovarian teratomas belong to the group of germ cell tumors. In fact, they are the most common benign ovarian neoplasms, representing 10 to 20% of these tumors.[1] They concern the reproductive age group.[3] From a clinical point of view, they can be asymptomatic or evoke abdominal complaints when they are large. Acute abdominal pain occurs in 5– 10% of all mature teratomas and is often due to ovarian torsion representing a serious complication.[6]

In addition, they are most often unilateral, but a bilateral presentation is very rare and represents only 10-15% of cases.[1,2] Imaging remains the mainstay in diagnosis of mature teratomas of the ovary and ultrasonography can be used as first line for initial evaluation.[1,2] The confirmation of the diagnosis will be made by histopathological examination.[4,7] The management of these tumors is essentially surgical and with torsion, it becomes a surgical emergency.[3,8] As bilateral presentation, these tumors with their rareness and the lack of protocol, their surgical approach is challenging, regarding the fertility and the iatrogenic menopause. There is no data in medical literature referring cases of mature ovarian teratomas in Haiti, even less of their bilateral presentation.

Here, authors present this rare case of a 42-years-old woman with bilateral mature ovarian teratoma, having an operative emergency with unilateral ovarian torsion in the context of Haiti where gang violence and political instability severely hinder access to healthcare, which are already struggling with resource and staff shortages. This leads to long delays and unsafe conditions for patients seeking standard care, that can worsen the management of this patient.

CASE REPORT

A 42-years-old female adult G2P2Lc2 came to the emergency department of Centre Medical de Frere with acute abdominal pain, ongoing vomiting for 48 hours. The patient previously went to a care facility where they treated

her for gastritis. Due to worsening symptoms, she was advised to seek further care and hoping for better management, but experienced a two-day delay in reaching our centre due to ongoing socio-political instability. She had no significant past medical, surgical or family history of chronic diseases, had her periods 2 weeks prior the symptoms the lasted 4 days with no significant changes of the length nor the abundance of bleeding. She did not use alcohol, tobacco or illicit drugs.

On physical examination, she had a fever of 38°C, a tachycardia up to 120 beats/mm the other vitals were witting the normal range, the patient was sweaty and appeared dehydrated. The abdomen was tendered and painful in the hypogastric region.

The vulva appeared to be filled with yellow discharges and the cervical exam was painful with the posterior fornix containing a firm mobile, regular masse. On the abdominal ultrasound we were able to visualize bilateral ovarian masses with similar sizes averaging 15×10 cm, solid, homogeneous, with thin wall, no increased vascularity.

Uterus, fallopian tubes were unremarkable (Figure 1). Authors were able to obtain some labs showing: Hg: 13,9 g/dL, WBC: 11,610 with neutrophiles at 86%, CRP: 48, BHCG: Negative, HIV: Negative, Syphilis: Negative. Considering the emergency of the case, we weren't able to have the tumor markers in pre-op and in post op we obtained them as described in Table 1.

After discussing the likelihood of "Ovarian torsion with patient having bilateral ovarian masses" was maintain as main diagnosis. Considering the patient had no further desire of pregnancy and the high suspicion of compromised ovarian tissue, then after counselling with the patient and her parents around the surgical menopause that would eventually occur, we have decided to proceed with total hysterectomy and bilateral salpingo-oophorectomy.

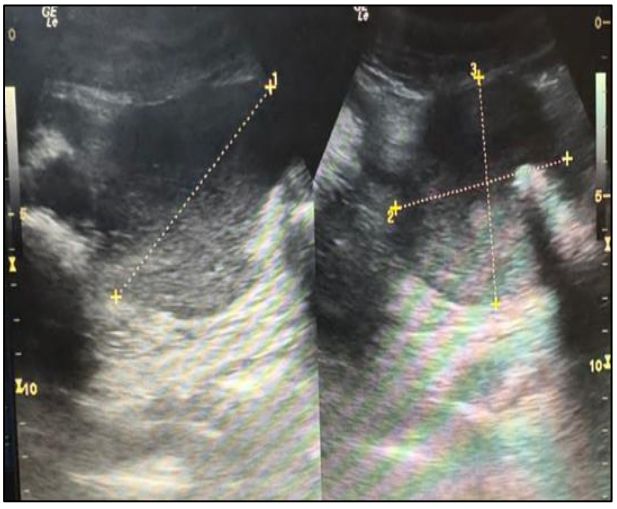

Figure 1: Visualization of the bilateral mass on ultrasound.

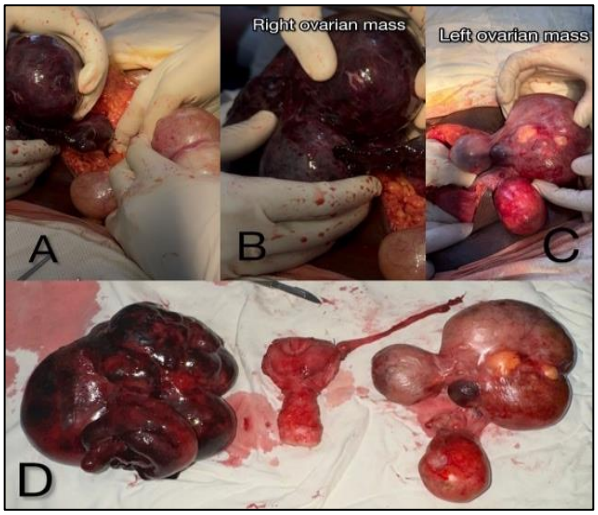

Figure 2: Operative findings (A) 2 Voluminous masses at laparotomy. B) Right ovarian large haemorrhagic tumor with torsion. C) Left ovarian multi-lobed cystic tumor. D) Operative part: bilateral adnexectomy and hysterectomy.

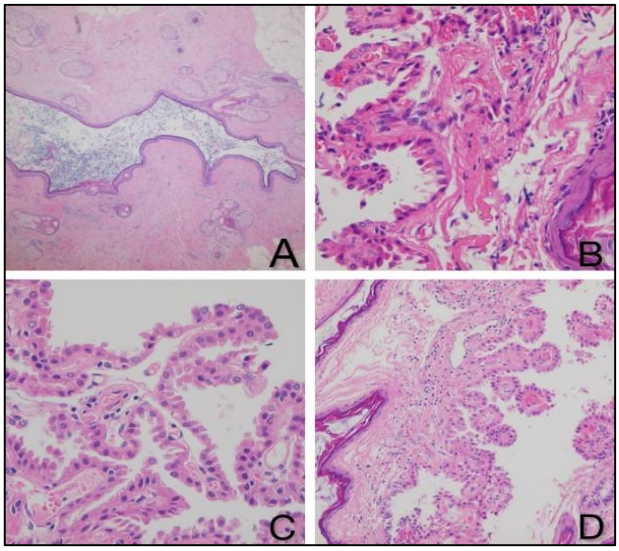

Figure 3 (A-D): Pathological slides of the tumor.

Before the laparotomy the patient received psychosocial support to prepare her for surgery and for the management of her symptoms post op. At the laparotomy we found 2 voluminous ovarian masses (Figure 2A). The right ovary was replaced by a necrotic solid mass that was non-adherent to the surrounding organs and exhibited two twits around the fallopian tube (Figure 2B). The left ovary was replaced a by a large pink mixed mass, multi-lobed with no adhesion with the surrounding organs (Figure 2C). No macroscopic abnormalities were detected in the uterus. No lymph node was palpated in the pelvic region. Appendices was normal. After advice in her post-operative follow-up and psychological consultation the patient was discharged 3th day post op without complications. The specimen was sent for histopathology study (Figure 2D).

The results were received 2 months later. On microscopy, we found tissues derived from ectoderm as skin with adnexa in almost all the cases, followed by keratin material, nervous tissue and nerve fibres, transitional epithelium. Also, from mesoderm as blood vessels, hematopoietic tissue, spindle shaped cells, adipose tissue, fibromuscular tissue, cartilage (Figure 3). Then, the pathologist diagnosed bilateral ovarian teratoma, with unilateral haemorrhagic infarction as torsion.

Table 1: Hormonal findings and tumors markers.

Tests	Unit	Results	Normal values
Ca 19.9	U/ml	164	0-36
Ca 125	U/ml	46,6	0-35
CEA	µg/l	2,70	0-5
LDH	U/l	-	150-450

DISCUSSION

The ovarian mature teratoma occurs in patients of reproductive age group 20-45 years predominantly.[5] Most patients with mature ovarian teratomas are asymptomatic unless complicated by torsion, but they can be revealed in 44% of cases by lower abdominal pain and 25% by abdominal mass or can be discovered incidentally in 21%.[3] In our context, the patient was 42 years old and symptomatic, with dull, aching abdominal pain and palpable mass, suggesting torsion. About 70% of ovarian torsion occurs on the right side, as seen in this case of bilateral mature ovarian teratoma during the laparotomy.[3] Often, these tumors have unilateral presentation, but bilateral finding is rare and remains challenging.[7]

As investigative procedure, pelvic ultrasound is typically the first-line imaging study for the initial approach of an adnexal mass, because it is non-invasive and accessible.[4,7] The patient had benefit it during the evaluation. The typical findings of these tumors at ultrasound may uniformly be hyperechoic or have bright linear to punctate echoes, also mature teratomas are detected as cystic or solid masses with fat or calcific differentiation in 90%.[6] Also colour doppler can help to find the twisted vascular pedicle regarding the torsion.[3,4]

Even on ultrasound we can have a variety of appearances, as in our case, we had bilateral ovarian mass, appeared solid, homogeneous and large.[3] At this time, other differential diagnosis of multiple complex ovarian masses includes combination of haemorrhagic cysts, endometrioma can be approached.[2] However, with the clinical presentation, the diagnosis of torsion on bilateral ovarian was considered and it became an emergency. Additional imaging such as CT or MRI, can helped in the characterization of ovarian mass, but may not be readily available in low-resource settings, complicating preoperative assessment.[7,9]

In early detection and characterization, these tumor markers such as CA125, CA19-9 and alpha-fetoprotein, LDH, can be used.[10,11] For, this type of tumor, serums markers levels was increased for Ca 125 and more often with Ca 19-9 and can be linked also to inflammation for this case. Occasionally, they help differentiate cystic adnexal masses.[10] In our emergency context, we made them in the immediate post-op for the follow up to define the type ovarian tumor and also to exclude any malignant transformation as immature teratoma, mucinous cystadenoma or cystadenocarcinoma of the ovaries or a metastatic disease. Only the 2 markers Ca 125 and Ca 19-9 were increased, as described below in the literature, but were not specific.

The management of these bilateral ovarian mature teratoma, remains challenging, because the rare incidence and also the lack of a standard protocol. However, surgical intervention is often performed and with the complication of unilateral torsion, it became a surgical emergency. The management depends on factors like age, fertility, the torsion and whether one or both the ovaries are involved.[9] In fact, Oophorectomy should be the standard operation in postmenopausal women and in perimenopausal women with multiple cysts in the same ovary or with large teratoma where there was no much ovarian tissue to conserve.[8]

As the rareness, 2 cases reviewed with the same age group 40-42, these patients had completed childbearing and would derive little benefit from retaining an ovary, hysterectomy with bilateral salpingo-oophorecctomy (BSO) was appropriate and done.[2,12] In our case, the patient, not desiring future fertility, had bilateral large masses with unilateral torsion and ovarian necrosis due to delayed evaluation. Therefore, we decided to perform also a total hysterectomy and bilateral adnexectomy. Also, she was informed about the occurrence of early menopause after surgery.[1,8] In this case, the resection was total, with no spills and histopathology noted mature teratoma. Per-op, certain approach such lymph node dissection or omentectomy was omitted, due the features of the mass intraoperatively, as any suspicious of malignancy.

On microscopy, histopathological examination revealed the mature teratomas consisted of tissues derived from ectoderm and mesoderm. With torsion, we can have extensive hemorrhage, congestion, dilated blood vessels and ischemia of mucosa and parenchyma.

For this case, we have findings related to both the ectoderm and mesoderm component in the bilateral mass, as hair follicles, sebaceous glands, keratin lamellae, adipose tissue and foci of dystrophic calcification, mature cartilaginous and nervous tissue, also in unilateral torsion necrotic haemorrhagic stroma was found. The histopathological results showed a bilateral ovarian mature teratoma. These tumors are mostly benign and malignant development occurs only in 0.2%.[1,2] We reinforced the counselling around the well prognostic and also the situation of surgical menopause. The post operatory follow-up was unremarkable after 4 months. This case

report highlights also how bad social political environment can worsen the clinical condition of a patient and hamper her access to health care and clinical follow-up especially in emergency.

CONCLUSION

Bilateral germinal ovarian tumor is very rare, can lead to diagnostic and management challenges, particularly when complicated by torsion. Given the patient's completed childbearing status and the large tumor size, including the torsion, total hysterectomy with bilateral salpingo-oophorectomy was an appropriate surgical approach. In low-resource settings with insecurity situation, delayed access to healthcare can significantly impact patient outcomes, that underlining the need for regular gynaecology follow-up which could improve the care of these patients, by early diagnosis and timely surgical intervention. Also, the development of radiological and histopathological diagnostic capabilities in such regions is crucial.

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