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A rare case of yolk cell carcinoma presenting as ovarian torsion

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Abstract
A Yolk Sac Tumour is the second most common malignant germ cell tumor and counts for only 1% of the malignant ovarian masses. It is most common in patients between 18 to 25 years. In most cases there are no clinical symptoms. However, abdominal swelling may be present in case of rapid growth. The size of tumors varies from 7 cm to 40 cm, with a median of 15-16 cm. The tumor is rarely bilateral (12-19%).
We report a case of 19 year old unmarried girl who came to our observation for an acute abdominal pain. Clinical examination evidenced a vague mass in the suprapubic region and a lower abdomen tenderness, the US imaging revealed a complex lesion of the right ovary (10 x 8 cm) and the alpha-fetoprotein (AFP) resulted high (>1000 ng/mL). The treatment consisted of unilateral salpingoophrectomy followed by 4 cycles of BEP protocol (Bleomycin, Etoposide, Cisplatin) but unfortunately patient died within 6 months as disease recurred in left ovary.

Keywords: Yolk sac tumor, germ cell tumor, ovarian torsion, ovarian tumor, alpha-fetoprotein

Introduction
Ovarian torsion results from partial or complete rotation of the ovary around its axis, leading to obstruction of the vascular pedicle [3, 4]. Although it occurs in normal ovaries, most frequently it is related to increased ovarian volume and benign masses [3-5].
Most ovarian masses in pediatric and adolescent patients are benign. A study of surgically managed ovarian masses in patients up to 19 years of age revealed that 2.3% were borderline and 5.3% were malignant, with no reported cases of ovarian torsion in either of these groups [6].
The rate of malignancy in ovarian torsion is low. When compared with malignant ovarian neoplasms, benign lesions have a 12.9-fold increased risk of being involved in adnexal torsion [7]. Diagnosis of ovarian torsion is challenging, since clinical features can be nonspecific and variable. The majority of patients present with lower quadrant pain, nausea and vomiting, which can mimic many other causes of abdominal pain [8, 9]. A palpable abdominal mass can also be present, a feature that is significantly associated with malignant masses [10].
Transvaginal sonography (TVS) is widely used for the evaluation of adnexal masses, and remains the primary diagnostic modality for suspected ovarian torsion [11, 12]. It is an accurate method for discriminating between benign and malignant lesions, especially when performed by an experienced ultrasonographer [13]. Advances in technology and the use of color Doppler have been helpful in the diagnosis of ovarian torsion, since they have increased diagnostic accuracy and positive predictive value [14].
Less frequent and unexpected findings sometimes appear together. Proper, accurate and complete preoperative evaluation is always essential, ensuring the best possible outcome for each patient. We report a case of ovarian torsion with a yolk cell ovarian tumor in a pubertal-aged girl.

Case Report
A 19-year-old, unmarried girl presented to the emergency room with localized hypogastric colicky pain that was started a week prior. Her menstrual cycles were regular and her last menstrual period was 2 weeks before admittance. She stated that she experienced intermittent episodes of low-level fever for the past 3 days. She did not use any drugs. No history of familial genetic diseases was reported by her and her parents. On physical examination, the patient was tachycardic (PR: 108) but was not febrile or hemodynamically unstable. Her abdominal examination revealed tenderness in the lower right quadrant and hypogastric area.

Laboratory tests including complete blood count (CBC), beta HCG, CA125, AFP, LDH, cea, CA19-9, urine analysis, and blood sugar (BS) as well as abdominal ultrasonography were requested with suspicion of ovarian torsion. The sonographic evaluation revealed a normal uterus and left ovary and a 90 × 80mm mass with heterogeneous echogenicity was observed with a high suspicion of right adnexa and ovarian torsion. Patient was immediately taken up for surgery.

She underwent a laparotomy. During the surgery with a infraumblical vertical incision, 150 mL of haemorrhagic coloured ascitic fluid was present, right ovarian mass of around 10*8 cm was present. Capsule got ruptured and was densely adherent to POD and gut. Adhesions were removed by blunt dissection. She underwent a right salpingo-oophorectomy (Figure 1 and 2). The samples from abdominal fluid and omentum were obtained for further analysis. In the abdominal exploration, no significant abnormalities were detected. The samples were sent for Histopathological examination. The histopathology report showed yolk cell carcinoma. The disease relapsed during chemotherapy and showed involvement of left ovary. Unfortunately, the general condition and symptoms made worse and she died 6 months after the initial diagnosis.

Discussions

A YST is the second most common malignant germ cell tumor and counts for only 1% of the malignant ovarian masses [1]. It is most common in patients between 18 to 25 years. In most cases there are no clinical symptoms. However, abdominal swelling may be present in case of rapid growth. The size of tumors varies from 7 cm to 40 cm, with a median of 15-16 cm. The tumor is rarely bilateral (12-19%). In symptomatic patients, abdominal pain is quite frequently complained leading to the discovery of the disease, as seen in our patient. Clinical signs consist of an abdominal or pelvic mass, vaginal bleeding, fever, ascites or peritonitis secondary to torsion, infection or rupture of the ovarian tumor [2]. The survival rate is higher in patients with earlier stage disease (stage I or II).

At present, the etiology of YST remains obscure. YST may develop from the false migration of embryonic primitive pluripotent germ cells or oncogenesis of the residual cells when they migrate from the genital ridge of the yolk sac endoderm [15]. These lesions typically determine abdominal pain with a rapidly enlarging abdominal mass [16]. Moreover, like in this case, ascites or peritonitis, due to the torsion of the lesion, are rare additional features [17]. CT scan and MRI are the most common techniques used to diagnose these lesions, but there are no specific signs to distinguish YST from other ovarian masses.

After treatment, periodic revaluations are required, such as abdominal and pelvic examination, CT scan, chest X-ray and AFP levels. YST are heterogeneous with a number of different histopathological subtypes. The typical histopathological features of YST are solid, tubular and focal papillary patterns with Schiller-Duval bodies and sinusoidal structures with fibrovascular cores lining formed by tumor cells, frequent mitotic figures and are cytokeratin positive [18]. In children, the vast majority of YST (85%) are in clinical stage I in comparison to a lower percentage (35%) in adults [19]. AFP can be applied as a feasible tumor marker because its level is elevated in > 90% of YST [20, 21]. In our case, the level of AFP was also increased (>1000 ng/mL).

Therapeutic approach is based on surgery and chemotherapy, depending on the stage of the tumor based on FIGO classification. The aim of surgery is removing the primary ovarian tumor without excessive morbidity. The standard surgical staging consists of peritoneal washing, peritoneal biopsies, and biopsies of any suspicious lesions. Factors related to good prognosis are no ascites at presentation, stage I disease, less than 42 days to AFP normalization, fertility-sparing surgery and a serum AFP half-life less of 10 days [22].

The BEP is considered the gold-standard first-line treatment for germ cell tumors at all stages [23]. Abdominal and pelvic examination, CT, chest X-ray and AFP levels are suggested during the follow-up [24]. Progressive or recurrent ovarian tumour after treatment with BEP chemotherapy has been reported to be associated with a poor prognosis [22]. The use of radiotherapy remains poorly defined because of its addiction to chemotherapy had no effect on improving survival.

In regards to the presented case, while our first suspicion was ovarian torsion, we also suspected that malignancy could cause such extensive destruction of ovaries. But unfortunately, the disease recurred in left ovay and patient died after 6 months.

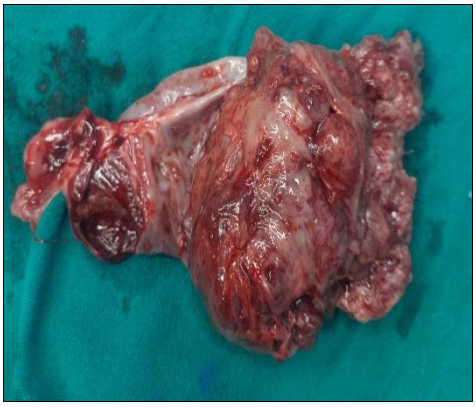

Fig 1: Resected specimen

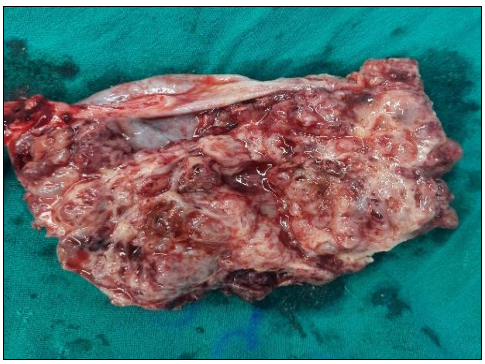

Fig 2: Resected specimen

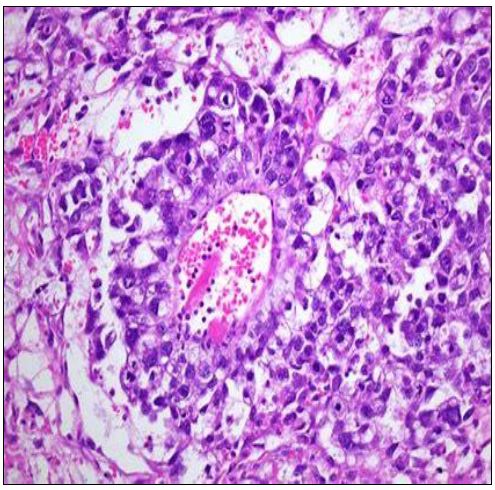

Fig 3: Microscopic view of YST

Conclusion

YST are rare neoplasms that usually occurs in the first two decades of life. Surgery with adjuvant chemotherapy is the standard management. It is important for clinicians to remember that patients with YST can present abdominal pain in an unusual way. Serum AFP is a useful marker for the diagnosis and management of YST. More research is required for investigating the mechanisms of pathogenesis in order to develop more effective treatments for YST.

Conflict of Interest

Not available

Financial Support

Not available

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