Internal hemorrhage caused by a twisted malignant ovarian dysgerminoma: Ultrasonographic findings of a rare case and review of the literature

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Summary

Purpose: Ovarian cancer presents as an acute abdomen very rarely. The purpose of the study is the description of a right ovarian malignant dysgerminoma presenting as an abdominal emergency.

Case: A 16-year-old white female presented with acute abdominal pain in the right iliac fossa. On physical examination the abdomen was acute and a mass in the right lower abdomen was palpated. The patient was sexually active and bimanual gynecological examination revealed the presence of a large lobulated solid tumor in the position of the right adnexa. Ultrasound examination showed the presence of a large, multilobulated, heterogeneous, predominantly solid pelvic mass. Color flow imaging showed intratumoral flow signals. The uterus and the left ovary had normal size and echo-texture. Fluid was found in the cul-de-sac and in Morisson's space. An immediate exploratory laparotomy exposed the presence of a twisted right ovarian mass and intraperitoneal hemorrhage. A superficial tumoral vessel actively bleeding was seen. Peritoneal fluid was obtained for cytology. The intra-abdominal hemorrhage ceased when the ovarian pedicle was clamped. The patient underwent right salpingo-oophorectomy and biopsy of the omentum. Pathologic analysis revealed a malignant dysgerminoma of the right ovary, expanding to the mesosalpinx. Cytology was positive for malignancy. Postoperative CT scan of the upper and lower abdomen was negative. The patient was assigned to FIGO Stage IIC and referred for platinum-based chemotherapy.

Conclusion: Ovarian malignant dysgerminoma may present as an acute abdomen because of torsion, passive blood congestion, rupture of superficial tumoral vessels and subsequent intra-abdominal hemorrhage. Ovarian dysgerminoma should be part of the differential diagnosis in a young woman with acute surgical abdomen and a solid heterogeneous pelvic mass detected by ultrasonographic scan.

Key words: Ovarian dysgerminoma; Ultrasonography; Color flow imaging; Abdominal emergency; Hemoperitoneum; Intra-abdominal hemorrhage; Torsion; Twisted; Ovarian mass.

Introduction

Adnexal torsion is an uncommon but significant cause of acute lower abdominal pain and morbidity in women. It is the fifth most common gynaecologic surgical emergency, with a prevalence of 2.7% [1]. Most cases of adnexal torsion are secondary to adnexal pathology [2] and it has been found that ovarian masses undergoing torsion are usually benign [3-6]. Hemoperitoneum due to a twisted solid ovarian mass is very unusual.

Dysgerminoma is a rare ovarian tumor, accounting for only 3-5% of all ovarian malignancies. It is a tumor of childhood and young adults, with about 90% of the tumors occurring in patients less than 30 years of age [7]. The clinical course of dysgerminoma is characterized by scanty local invasiveness, early lymphatic spread and rarely hematogenous dissemination [8]. Another important issue for ovarian dysgerminoma is related to the high incidence of bilateral disease (10-20%) [8, 9]. The clinical presentation of ovarian dysgerminoma is non-specific

as for ovarian carcinomas. The women may complain of an abdominal mass, dysmenorrhea, dyspepsia and abdominal discomfort [8]. Acute abdomen due to hemoperitoneum secondary to ovarian dysgerminoma is extremely rare [10, 11]. Often, the tumor is discovered during a routine examination [8]. On imaging studies, ovarian dysgerminomas have been characterized as non-specific solid masses [12].

In this study, we report a case of a right ovarian malignant dysgerminoma presenting as an abdominal emergency. Also, the interesting ultrasonographic findings are presented and the international literature is reviewed.

Case Report

A 16-year-old, white, sexually active nulligravid young woman presented to the emergency room complaining of sudden onset right lower quadrant abdominal pain. The pain had started eight hours before admission with increasing intensity and become very sharp during the last two hours. The patient was nauseous and had vomited a number of times. Body temperature was normal. She denied chills, dysuria, constipation, diarrhea and vaginal discharge. She denied any recent menstrual
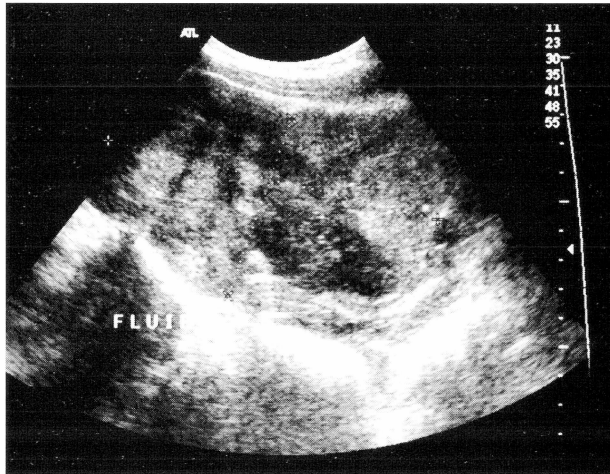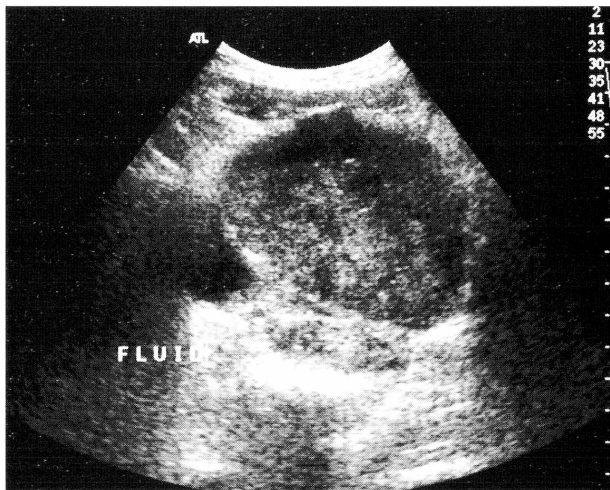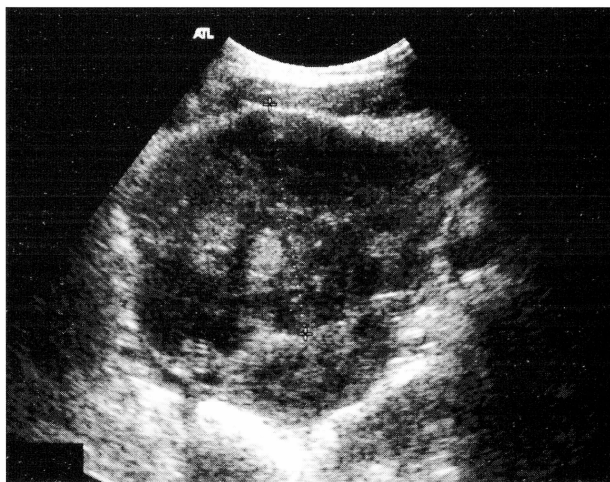
Fig. 1

Fig. 2

Fig. 3

Figures 1, 2, 3. — Transabdominal ultrasound scan showing a large, multilobulated, heterogeneous, predominantly solid pelvic mass. The dimension of the tumor was found to be 11.68 x 6.48 x 6.93 cm. The bladder was displaced leftward and the uterus posteriorly. Fluid in the cul-de-sac with low levels of echogenic debris was found. Surgery demonstrated the presence of hemoperitoneum of about 500 ml with recent blood clots.

irregularity and was on day 35 of a 28-day menstrual cycle. Her past medical history was uneventful. On physical examination her lower abdomen was markedly tender mainly in the right lower quadrant, with moderate spasm and rebound tenderness in the right iliac fossa. A large mass was noted to occupy the lower right abdomen and pelvis. Peristaltic sounds were diminished. A large lobulated solid tumor in the position of the right adnexa and in front of the uterus was felt during bimanual gynecological examination. The cervix was closed, firm and very tender on motion. The patient was hemodynamically stable. The review of her systems was otherwise entirely normal. Hemoglobin concentration was 10.4g/dl, hematocrit 32.1% and white blood count 12,000 cells/ml with 67.8% polymorphonuclears. Ultrasonographic examination demonstrated the presence of a large, multilodulated, heterogeneous, predominantly solid mass. The tumor measured 12 x 6.48 cm and expelled the bladder leftward and the uterus backward. Fluid was found in the cul-de-sac with low levels of echogenic debris (Figures 1, 2, 3). Additional color flow imaging showed intratumoral vascularization (Figure 4). The right kidney was observed with mild hydronephrosis. The uterus and the left ovary had normal size and echo-texture. Also, fluid in Morisson's space was detected. Serum levels of α-fetoprotein (αFP), lactate dehydrogenase (LDH), CEA and Ca19-9 were within normal values. Serum β-human chorionic gonadotropin (β-hCG) was negative. Serum levels of Ca-125 were elevated (42 IU/ml) (normal, less than 35 IU/ml). Urinalysis and liver function tests were within normal limits. Clotting time and bleeding time were within normal limits; platelets appeared adequate in number and quality on a blood smear. Serum calcium levels were normal. Chest X-ray examination was negative.

Because of the acute abdomen an immediate exploratory laparotomy was performed. A vertical, midline, supra-infra-umbilical incision exposed the presence of a twisted right ovarian mass and hemoperitoneum of about 500 ml with recent blood clots. A superficial vessel actively bleeding was seen in the wall of the tumor; the intra-abdominal hemorrhage ceased when the ovarian pedicle was clamped. No other source of bleeding was encountered. Peritoneal fluid was obtained for cytology. The patient underwent right salpingo-oophorectomy and biopsy of the omentum. The abdominal cavity was irrigated and carefully inspected, without encountering any visible findings suspicious of extra-ovarian malignancy. The uterus, left ovary, left fallopian tube and appendix were found to be normal. No enlarged pelvic or para-aortic lymph nodes were found. Frozen sections of the ovarian tumor suggested the possibility of a tumor originating from the germ cells with moderate cellular atypia. As the left ovary was normal in size, shape and consistency, no wedge biopsy was taken from it. No drainage was installed, but a nasogastric tube was placed. Blood tranfusion was not given. The postoperative course of the patient was uneventful.

Postoperatively, CT scan of the upper and lower abdomen was negative. Figure 5 shows the gross appearance of the ovarian dysgerminoma. The tumor was solid and multilobulated and it measured 13.5 x 7 x 8 cm. The tumor retained the general configuration of the ovary and had a "brain-like" appearance. Pathologic analysis of the specimen revealed a malignant dysgerminoma of the right ovary which expanded to the mesosalpinx. Extensive necrotic areas of the neoplastic tissue were observed with oedema and congestive blood vessels. The stained cytologic smears of the peritoneal fluid were positive for malignancy. The tumor was staged as IIc according to the guidelines of the International Federation of Gynecologists and Obstetricians (FIGO) classification system for ovarian cancer [13] and the patient was referred for platinum-based chemotherapy according to Dimopoulos et al. [14].
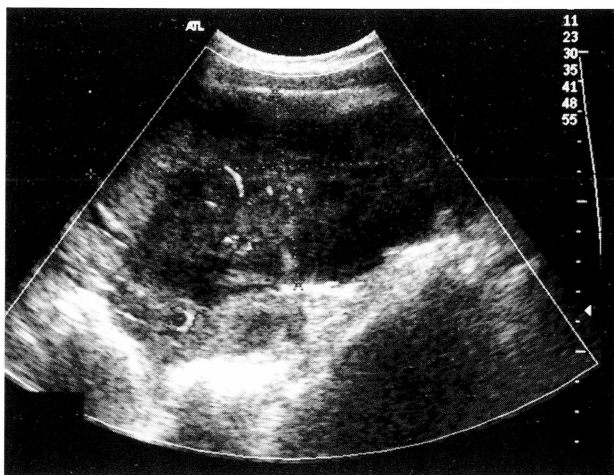
Figure 4. — Color flow imaging showing intratumoral vascularization, although immediate exploratory laparotomy exposed the presence of a twisted right ovarian mass.
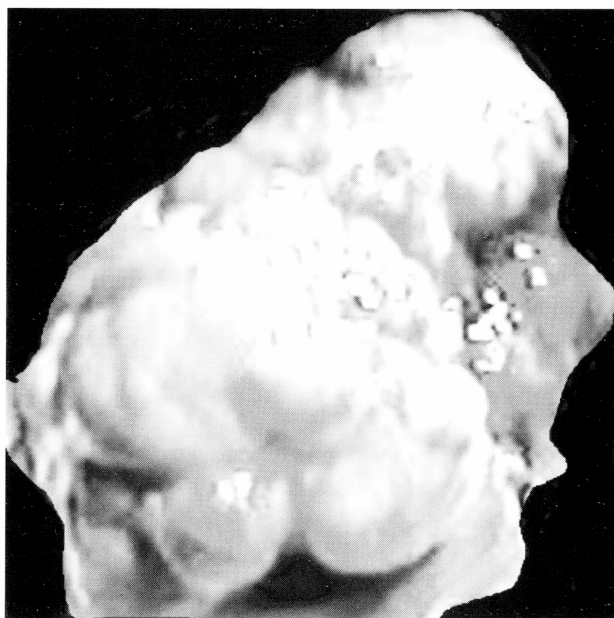

Figure 5. — Gross appearance of malignant dysgerminoma of the right ovary: the tumor is solid and multilobulated. The tumor retains the general configuration of the ovary and gives a "brain-like" appearance.

Discussion

Approximately 5% of malignant ovarian tumors are germ cell tumors. Malignant ovarian germ cell tumors can be divided into two main groups: (1) dysgerminomas and (2) non-dysgerminomas (immature teratomas, endodermal sinus tumors, mixed germ cell tumors, embryonal carcinomas, polyembryomas and choriocarcinomas) [15]. These tumors occur primarily in young women with a median age at diagnosis of 16 to 20 years [16]. Ovarian dysgerminoma is analogous to seminoma of the testes [17] and is most often diagnosed at an early stage. In a

review of 271 cases of ovarian dysgermimoma, 70% were Stage I at the time of surgery [17]. Ovarian dysgerminoma is composed of primordial germ cells that are not differentiated into embryonic or extraembryonic structures. The concept of origination from primordial germ cells is strengthened by the occurrence of homologous neoplasms in the testes and along the route of migration of the primordial germ cells from the yolk sac of the embryo to the primative gonad [17].

Signs and symptoms of dysgerminoma are not different from ovarian tumors of other histologies [18]. Hemoperitoneum resulting from a solid ovarian mass, just like the ovarian dysgerminoma, is extremely rare [11]. In our case, the torsion of the ovarian dysgerminoma seemed to cause passive blood congestion, subsequent rupture of a superficial tumoral vessel and spontaneous internal bleeding. The work-up of ovarian dysgerminoma should include chest X-ray, routine blood tests, tumor markers, ultrasonography and CT scan [8]. In addition to that, serum calcium levels should be checked in all children with solid ovarian tumors. Hypercalcemia in such circumstances may be resistant to medical management and surgical resection of the tumor results in the normalization of calcium levels [19]. Ovarian tumors markers are typically not expressed in most ovarian dysgerminomas; however, 5% of ovarian dygerminomas are found to contain syncytiotrophoblastic cells and in these cases are associated with the production of β-human chorionic gonadotrophin (β-hCG) [20, 21]. Elevation of serum lactate dehydrogenase (LDH) has also been noted in some cases of ovarian dysgerminomas [22]. Also, abnormal liver function tests may be elevated up to two-fold that the normal cut-off value, and when associated with markedly elevated LDH, this finding is characteristic of the presence of an ovarian dysgerminoma [15]. Ca-125 is a tumor marker specific to epithelial ovarian tumors. AFP is strongly related to yolk sac tumors and embryonal carcinomas and β-hCG to choriocarcinomas. If serum levels of either LDH or hCG are elevated preoperatively, serial determinations should be performed after therapy to monitor disease status [17]. In our case, although surgery was performed on an emergent basis, serum was obtained preoperatively, stored and the serum levels of tumor markers were analyzed when the patient was confirmed to have ovarian dysgerminoma. The serum levels of tumor markers and lactate dehydrogenase were within normal limits, apart from the serum levels of CA-125 and the reason for this elevation remains unknown. Also, Tanaka *et al.*, reported a case of ovarian dysgerminoma with increased CA-125 levels [18]. Radiologic findings of ovarian dysgerminoma have not been well described. Tanaka *et al.* [18] and Kim and Kang [12], reported the CT and MRI findings of ovarian dysgerminoma in six cases. They found that the tumor had a multilobulated solid appearance with the lobules divided by fibrovascular septa [12]. In addition, Doppler sonography demonstrated the presence of prominent arterial flow within the fibrovascular septa with RI ranging between 0.44 and 0.70 and PI between 0.60 and 1.32 [12]. In our case, the

tumor was a heterogeneous predominantly solid mass with a multilobulated appearance. The color flow imaging demonstrated intratumoral blood flow, although the right adnexae was twisted. This finding is explained by the degree of the vascular compromise. As is known, ovarian torsion initially interferes with the venous and lymphatic circulation and, if unrelieved, progresses to occlusion of the arterial circulation [2]. Therefore, persistent arterial flow cannot rule out the diagnosis of early or incomplete torsion of the ovary. The specific indication for complete occlusion of both venous and arterial vessels is the absence of flow to the ovary detected by color Doppler ultrasonography [23]. In general, solid ovarian masses in children are much more likely to be malignant as compared with either complex or simple cysts [2]. The differential diagnosis of a solid ovarian mass in a young woman detected utrasonographically includes a fibroma, a fibrothecoma or a dysgerminoma [15].

The management of dysgerminoma includes surgery, radiation and chemotherapy. The treatment must be tailored to the woman's individual needs: preserving childbearing is an important issue as this disease especially affects young women. The rarity of the disease and the limited number of patients included in the published series have led to some controversies regarding the best management [8]. The issue of conservative versus nonconservative primary surgery in patients with dysgerminoma has been vigorously debated for decades. Proponents of non-conservative therapy in the past have used the following arguments: (i) 10 to 20% of patients with ovarian dysgerminoma have bilateral ovarian disease. (ii) 16% to 52% of patients with ovarian dysgerminomas were found to have a recurrence of their tumor, with up to 35% of the recurrence occurring in the conserved contralateral ovary. (iii) Survival after recurrence seems to be only 45-65% [8, 9, 24-29]. In contrast, several characteristics of this tumor make conservative therapy with preservation of reproductive capacity extremely attractive: (i) The mean age of occurrence of this tumor varies between 18 and 23 years of age. (ii) About 90% of the patients are younger than 30. (iii) The tumor is confined to a single ovary in approximately 80-90% of patients. (iv) The tumor displays exquisite sensitivity to platin-based chemotherapy and radiation therapy in both adjuvant and salvage treatment settings [8, 24, 26, 30-40]. Casey *et al.*, in a retrospective study examined how conservative and non-conservative primary surgery affected survival and patterns of disease recurrence. The authors found that survival was similar both in the patients who underwent conservative primary surgery (93%) and in those who underwent non-conservative primary surgery (90%) [24]. The progress in chemotherapeutic regimens and a better knowledge of the natural history of ovarian dysgerminoma have limited the role of radiation. Chemotherapy offers the advantage of allowing preservation of ovarian function and the woman's fertility. On the other hand, the side-effects of chemotherapy are many and vary depending on the drugs used, while late injuries due to radio-

therapy are seldom reported if the dose delivered is kept within the tolerance limits of the small bowel [8]. Some authors suggested that in patients with large ovarian dysgerminomas (> 10 cm) there is a greater chance of recurrence, and therefore adjuvant therapy should be given. However, most authors agree that tumor size is not prognostically important and that these patients do not require additional therapy [41].

In our case, the patient underwent the operation in the Gynecological Department of a General Hospital, because of the urgency for surgery; the ovarian dysgerminoma presented as an abdominal emergency. It has been published that more than 80% of women with ovarian germ cell tumors will undergo the initial operation in general gynecological institutes [42]. Our patient underwent right salpingo-oophorectomy and biopsy of the omentum. We obtained peritoneal fluid for cytological examination, inspected the entire peritoneal cavity without encountering any visible findings suspicious of extra-ovarian malignancy. The gross appearance of the tumor was similar to the description of other authors [43]. Intra-operative frozen sections of the ovarian tumor suggested the possibility of a tumor originating from the germ cells with moderate cellular atypia. We did not perform wedge resection of the contralateral ovary because it was normal in size, shape and consistency. Generally, the reason not to perform a wedge resection of a normal contralateral ovary at the initial operation is related to concerns regarding fertility following wedge resection of an ovary, as any operative procedure may be associated with a subsequent adhesion formation and tubo-peritoneal disease [15]. Also, oligo-ovulation or anovulation may result after post resection of the contralateral ovary [44]. Disaia and Creasman reported that their routine is not to wedge or bivalve the opposite ovary if it is normal in size, shape and consistency [41]. In our case, the FIGO stage was assigned as IIC and the patient was referred for platinum-based chemotherapy [14]. As we mentioned above, chemotherapy is preferred in patients who desire to maintain their fertility. In patients with ovarian dysgerminoma, FIGO Stage IA, unilateral salpingo-oophorectomy is sufficient. In patients with bilateral ovarian dysperminomas (Stage IB) some researchers have attempted preservation of the least affected gonad followed by chemotherapy, with no adverse outcome and fertility preserved [45]. Stage IC tumours are treated with conservative unilateral salpingo-oophorectomy followed by chemotherapy [16]. If metastatic disease is noted on the uterine serosa in women desiring future fertility, a hysterectomy may be avoided due to the chemosensitivity of dysgerminomas, and local excision with preservation of the corpus should be considered in some selected individuals [16]. Conservative surgery for patients with FIGO Stage III and IV dysgerminomas involves unilateral salpingo-oophorectomy and cytoreductive operation plus chemotherapy, followed by a second cytoreductive operation [16].

In contrast to surface spread, which is typical of epithelial ovarian tumors, ovarian dysgerminoma has a propen-

sity for early lymphatic spread to the pelvic, para-aortic, mediastinal and supraclavicular lymph nodes [9]. Current 5-year survival rates for this tumor approach 90% for Stage IA and 63-83% for more advanced disease [24, 30, 32, 33, 35, 36]. However, 2-year survival without recurrence indicates an excellent chance of cure [46], because approximately 90% of recurrences will appear in the first two years after initial therapy [41]. Late relapse may rarely occur in patients with Stage IA ovarian dysgerminoma [47, 48]. Fortunately, the majority of recurrences can be successfully eradicated by radiation therapy or chemotherapy [41].

About 5% of ovarian dysgerminomas are associated with chromosomal anomalies [49, 9]. Therefore, in cases of a prepubertal pelvic mass, karyotype should be carried out. If the results show gonadal dysgenesis and the presence of a Y chromosome, both gonads should be removed since 25-50% of these cases will develop ovarian malignancy, usually gonadoblastoma or dysgerminoma [9, 24]. In these cases, the uterus should be left *in situ* for future embryo transfer [9].

In conclusion, we presented an unusual case of internal bleeding associated with torsion of the pedicle of a right ovarian dysgerminoma and subsequent acute intra-abdominal crisis, which led to the diagnosis of the tumor. The interesting ultrasonographic findings of the ovarian dysgerminoma were also presented in order for obstetrician-gynecologists to keep in mind the differential diagnosis of ovarian dysgerminoma when a heterogeneous, predominantly solid pelvic mass is detected ultrasonographically in young patients.

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