Case Report

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Ovarian Dysgerminoma with Torsion: A Case Report

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Dysgerminoma is one of three most common ovarian tumors and accounts for 2% of ovarian neoplasms. Dysgerminoma usually affects adolescents and young adults. Dysgerminoma can be accompanied with elevated serum levels of lactate dehydrogenase (LDH) and placental alkaline phosphatase (PLAP). Radiological diagnosis of dysgerminoma is difficult due to its non-specific features. Clinically, the diagnosis is usually confirmed by tissue expression of CD117, D2-40 and OCT4. A 50-year-old woman suffering from persistent lower abdominal dull pain radiating to right flank region visited our emergency room. A palpable firm mass located at right lower quadrant of abdomen was noticed during physical examination. Gynecological sonography and computerized tomography revealed a left adnexal mass with heterogeneous solid components. Serum β-hCG level was 5,823.7 mIU/mL. Laparoscopic intervention was carried out under the impression of left ovarian cystic teratoma superimposed with ectopic pregnancy. Laparoscopic right salpingo-oophorectomy was performed to remove a necrotic right ovarian mass with hydrosalpinx and torsion. Pathological examination showed dysgerminoma with torsion. FIGO stage was suggested to be Ic1 (T1c1NxMx). No tubal pregnancy was found. Hysterectomy, left salpingo-oophorectomy, pelvic lymphadectomy and partial omentectomy were performed during further staging surgery. Four cycles of BEP (bleomycin, etoposide, cisplatin) adjuvant chemotherapy were implemented. In the subsequent follow-up at outpatient clinics with CT scan, no tumor recurrence has been noted.

Key words: dysgerminoma, teratoma, ectopic pregnancy, immunohistochemistry

Case Report

A 50-year-old patient, gravida 2, para 2 (normal spontaneous vaginal delivery), with last menstrual period on March 6, 2020 presented to our emergency room with lower abdominal pain for 3 days. She has suffered from irregular menstrual cycle since November, 2019. Her previous menstruation was recorded on January 31, 2020. The persistent dull pain radiated to right flank region. Physical examination found a palpable firm mass located at right lower quadrant of abdomen. Pregnancy test was positive with serum β-hCG level at 5,823.7 mIU/mL. Gynecological sonog-

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raphy revealed a left adnexal mass 10.4 cm × 7.9 cm in size with heterogeneous solid components (Fig. 1A). No intrauterine pregnancy was noted. No fluid accumulation was found in the cul-de-sac. Pelvic examination found that the cervix was intact. Copious clear vaginal discharge without odor was also observed. According to the patient, she has suffered from increase of both clear vaginal discharge and sexual libido since November, 2019. Computerized tomography (CT) scan demonstrated a fat-containing lesion about 13 cm in diameter in left ovary (Fig. 1B) accompanied with bilateral pleural effusion (Fig. 1C). Thus, left ovarian cystic teratoma superimposed with ectopic pregnancy was suggested. Small amount of blood was found to accumulate in the cul-de-sac (Fig. 1D). Laparoscopic exploration was performed under the impression of left adnexal tumor with torsion or hematoma-embedded tubal pregnancy. During the surgery, inconsistent to radiological findings, a necrotic right ovarian mass with hydrosalpinx and torsion (Fig. 2A & 2B) was found. No active bleeding was found. Thus, laparoscopic right salpingo-oophorectomy was carried out. Pathological examination showed a dysgerminoma with torsion FIGO stage was suggested to be Ic1 (T1c1NxMx). No tubal pregnancy was found.

H & E stain showed uniform cells separated by fibrous septa with clusters of lymphocytes (Fig. 3A). Cells with mitotic figures were also found (Fig. 3B). Immunohistochemistry (IHC) displayed positive membranous and cytoplasmic staining of CD117 and D2-40 (Fig. 3C & 3D). OCT3/4 was found to be positive in nucleus and cytoplasm (Fig. 3E). Sal-like protein 4 (SALL4) and placental alkaline phosphatase (PLAP) were focal positive in nucleus and focal weakly positive in cytoplasm, respec-

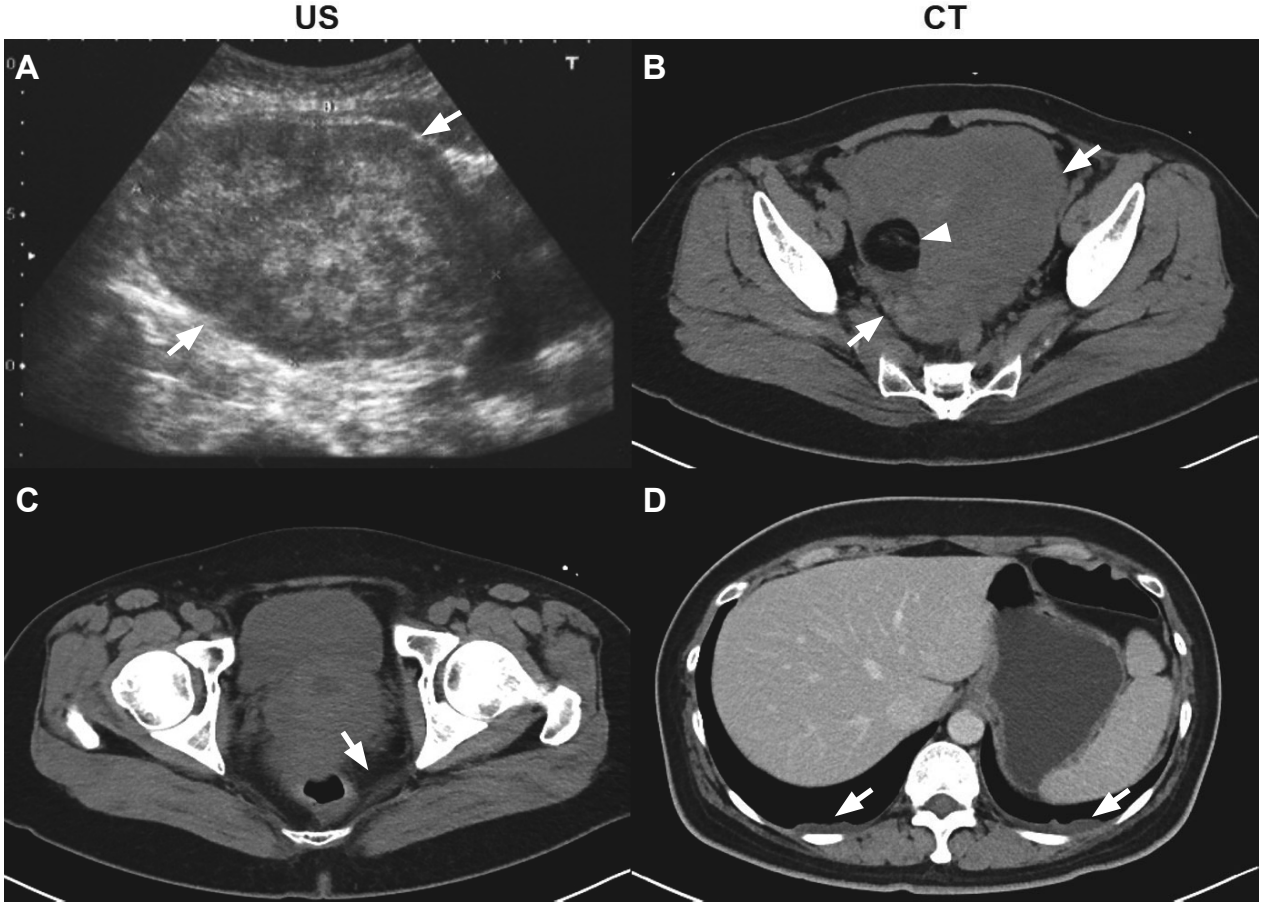

Fig. 1 Imagery results of the patient. (A) Transabdominal sonography revealed a left ovarian tumor with heterogeneous contents (arrows). CT scan demonstrated (B) a pelvic tumor (arrows) with fat component (arrowhead) accompanied by (C) hemoperitoneum (arrow) and small amount of pleural effusion (arrows).

tively (Fig. 3F & 3G). CD30 was not detected (Fig. 3H). The serum levels of tumor markers, including CA125, CA199, alpha-fetal protein (AFP) and lactate dehydrogenase (LDH) were 106.5 U/mL, 52.71 U/mL, 2.52 ng/mL and 885 U/L, respectively. One day following surgery, β-HCG level was decreased to 393.5 mIU/mL. Six weeks after surgery, the levels of CA125 and CA199 were decreased to 9.3 U/mL and 4.32 U/mL, respectively.

Further staging surgery with hysterectomy, left salpingo-oophorectomy, pelvic lymphadectomy and partial omentectomy was then carried out 10 days after the first laparoscopic surgery. No distant metastasis was found by a thorough survey of other organ systems. Four cycles of adjuvant chemotherapy using BEP (bleomycin, etoposide, cisplatin) regimen were administered. No tumor recurrence was found by CT scan in the subsequent follow-up at outpatient clinics in the past 10 months.

Discussion

Dysgerminoma is the second most common malignant ovarian germ cell tumor,

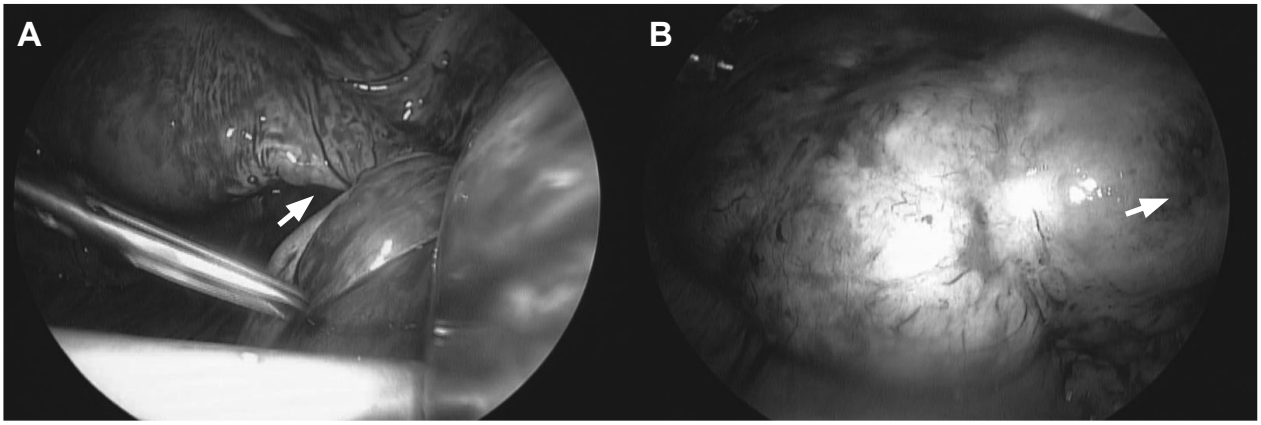

Fig. 2 Observations during laparoscopy. (A) A right ovarian solid tumor with torsion (arrow) and (B) necrotic changes (arrows) was found.

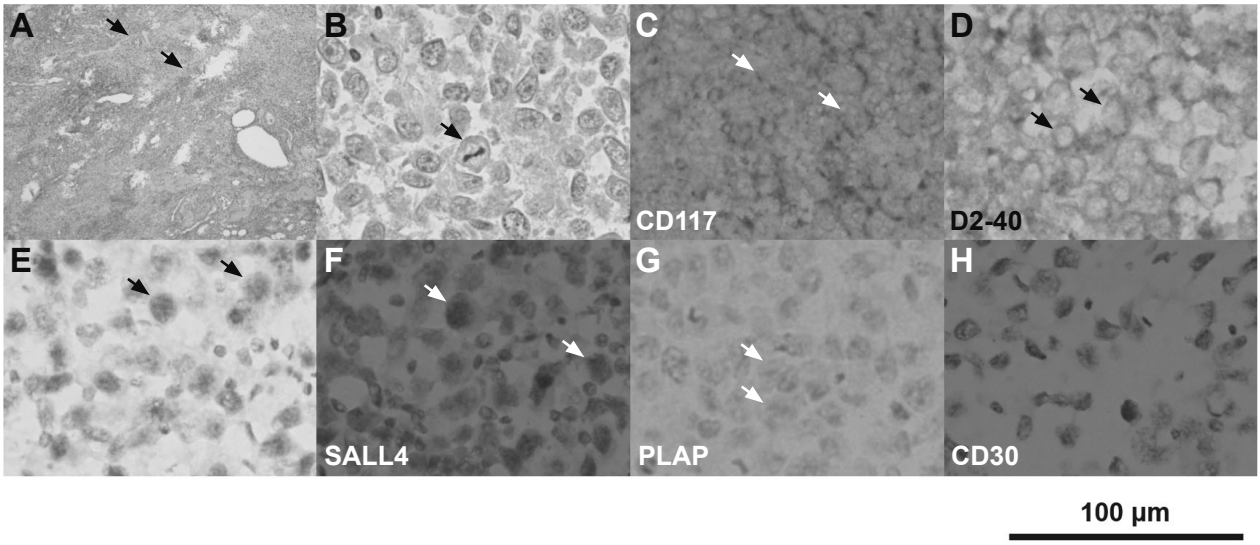

Fig. 3 Pathological examination. (A) H & E stain demonstrated nests of squared-off tumor cells were separated by fibrous septae with lymphocyte infiltration (arrow). (B) Cells with mitotic features (arrows) were also found. (C) CD117 and (D) D2-40 were found to be positive in membrane and cytoplasm (arrows). (E) OCT3/4 is expressed in nucleus and cytoplasm (arrows). Focal positive staining was found in nucleus and cytoplasm for (F) SALL4 and (G) PLAP, respectively (arrows). The tumor tissue did not express (H) CD30.

which is one of three most common ovarian tumors. It accounts for 2% of ovarian neoplasms. Dysgerminoma usually affects reproductive age women, particularly adolescents and young adults. Diagnosis in perimenopausal age is less common. Dysgerminoma can be accompanied with elevated serum levels of LDH and PLAP produced by syncytiotrophoblastic giant cells detected in some cases. Although only 3 – 5% of dysgerminomas produce β-hCG, β-hCG levels were found to be increased in this case, suggesting the existence of syncytiotrophoblasts in this patient. Serum levels of PLAP were not tested in the current patient. The elevated β-hCG levels and non-specific radiological features led to the misinterpretation of CT scan. Early tumor recurrences can be monitored by serum LDH, PLAP, and β-hCG levels.[1] Unless the tumor contains mixed components of germ cell tumor and a yolk sac element, the AFP levels will not be elevated. Although small amount of pleural effusion was detected by CT scan, a thorough survey did not find any distant metastasis in this patient. Thus, the cause of pleural effusion still needs to be determined.

IHC plays a pivotal role in characterizing germ cell tumors. Immunochemical profiles are usually informative in differentiating various ovarian tumors. Specifically, dysgerminoma expresses CD117, D2-40 and OCT4.[2] Yolk sac tumor can be distinguished by AFP and glypican-3. OCT4, CD30 and SOC2 are used to detect embryonal carcinoma. Sex cord tumor is characterized by calrectinin, inhibin, SF-1 and FOXL2. Steroid tumor is positive for melan-A.[3] PLAP is for malignant germ cell differentiation. The diagnosis of the current patient was confirmed by positive staining of CD117, D2-40 and OCT4. Expression of SALL4 confirms the malignant germ cell differentiation[4] and metastasis.[5] Both OCT3/4 and SALL4 are also used to identify the pluripotency of the tumor.[6]

The patients with dysgerminoma usually present with non-specific symptoms, such as pelvic fullness, lower abdominal pain, early satiety, urinary frequency and dysuria. The etiology of dysgerminomas is thought to be escape of germ cells from primordial follicle without normal contact inhibition. Like other seminomatous germ cell tumors, dysgerminomas are very sensitive to bother radiotherapy and chemotherapy. Thus, the long-term survival rate for the patients is perfect.[7]

In summary, the diagnosis of the current patient was confirmed by pathological examination with immunohistochemical staining of various markers associated with dysgerminomas. The initial diagnosis was misled by elevated serum β-hCG levels and non-specific radiological features. Since gonadoblastomas may give rise to dysgerminomas, a thorough sampling is required to rule out its presence.

Author Contributions

Shu-Jiin Tey: Conception and design of study, Acquisition of data, Drafting the manuscript, Approval of the version of the manuscript; S. Joseph Huang: Drafting the manuscript, Approval of the version of the manuscript; Chih-Chen Chen: Conception and design of study, Acquisition of data, Drafting the manuscript, Approval of the version of the manuscript.

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Data Availability Statement

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Conflict of Interest

The authors declare no conflict of interest.

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