中文題目: 一位腹膜透析病患在卵巢發生成人顆粒細胞瘤。

英文題目: Adult granulosa cell tumor in the ovary of a peritoneal dialysis patient

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Abstract

An adult granulosa cell tumor is a rare ovarian cancer that has never been reported in dialysis patients. We present here an unusual case of on and off hemoperitoneum for one month in a female peritoneal dialysis patient. Transabdominal sonography during a follow-up for a cystic mass in the right ovary showed enlargement, so we operated to remove the mass. The pathology showed a stage I granulosa cell tumor, enlightening us to the fact that unusual patterns of bloody ascites in peritoneal patients should be carefully examined.

Case report

Adult granulosa cell tumors originate from sex cord-stromal cells, with uterine bleeding being the most common initial presentation. Menstruating women on peritoneal dialysis often experience bloody ascites, so it is easy to overlook malignancy in this group of patients.

Our patient, a 23-year-old woman, had started peritoneal dialysis in 2003 due to kidney agenesis and presented with intermittent bloody ascites for one month beginning October 2010. These bloody ascites sometimes occurred during her menstruation, but also showed up intermittently at other times. Her menses were irregular. When she visited her gynecologist, a right ovarian cystic mass of approximately 5cm was noted. She was followed up regularly by the gynecologist, who determined that the mass was steadily enlarging. The ovarian mass located via transabdominal sonography measured 6.2 cm on 1/18/2011. A CT with contrast was done on 2/27 and also showed a right ovarian cystic mass (Figure 1). Additional ultrasounds showed increases in size to 8.7 cm on 3/18, and 10.5 × 9.3 × 9 cm on 4/19 (Figure 2). The ascites cytology produced negative results. Her estradiol level was 2992.6 pg/ml. We shifted her from peritoneal dialysis to hemodialysis and operated to excise this right ovarian cystic mass. The pathology showed an adult granulosa cell tumor (Figure 3) with some Call-Exner bodies. Her lymph nodes were all negative (0/7). She remained on regular hemodialysis with a plan to return to peritoneal dialysis in one month. Her peritoneal dialysis is now running smoothly.

Hemoperitoneum is a common complication of peritoneal dialysis. The etiology could be related to the catheter, obstetric and gynecologic conditions, intra-abdominal organs, vascular problems, earlier procedures, bleeding diathesis, infection, or retroperitoneal hematoma (1). More than half of menstruating women on peritoneal dialysis may experience hemoperitoneum at least once, either during their period or 1–2 weeks prior (2). Bloody ascites have often been reported under the circumstances of menstruation, ovulation, ovarian cyst rupture and pregnancy. To our knowledge, an adult granulosa cell tumor has not been previously reported in a peritoneal dialysis patient.

Hemoperitoneum has been reported with a hepatoma rupture (3) or after a transcatheter arterial chemoembolization for hepatoma in peritoneal dialysis patients. However, granulosa cell tumors have never been mentioned. Our patient had negative cytology findings. The hemoperitoneum may have been due to her high estradiol level stimulating her ovaries.

Adult granulosa cell tumors account for 4.3% of ovarian malignant neoplasms. The most common presenting symptom has been abnormal uterine bleeding (53.7%). The initial stage seems to be the single most important prognostic factor in ovarian granulosa cell tumors (4). Their tendency to recur years after the initial diagnosis is their most prominent characteristic. The secretion of estradiol is the reason for signs at presentation such as vaginal bleeding in women or precocious puberty in girls. Abdominal pain and hemoperitoneum, which can occur occasionally, are attributable to tumor rupture. Inhibin B and anti-müllerian hormone (AMH) are both useful serum markers for diagnosis and especially for the follow-up of patients with a granulosa cell tumor (6). Mutant FOXL2 (a missense point mutation, 402C→G (C134W)) is highly associated with adult granulosa cell tumors and has been postulated as a potential driver in the pathogenesis of adult-type granulosa cell tumors (7).

In our peritoneal dialysis patient, a Tenckhoff catheter opened a window to the peritoneum. Hemoperitoneum can be detected earlier than vaginal bleeding or other signs of tumor. Uremic toxins may hinder the effects of estradiol, explaining why our patient did not present with vaginal bleeding.

In conclusion, we wanted to draw your attention to this case of unusual bloody ascites in a woman undergoing peritoneal dialysis, which proved to be an adult granulosa cell tumor in her right ovary. The bloody ascites did not signal peritoneal metastasis, leading us to conclude that bloody ascites could be the initial presentation

of ovarian cancer in peritoneal dialysis patients instead of vaginal bleeding. Clinicians need to pay close attention to any unusual patterns of bloody ascites in peritoneal dialysis patients.

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Figure 1. CT of ovarian tumor (right-facing arrow) and the Tenckhoff catheter (downward arrow).

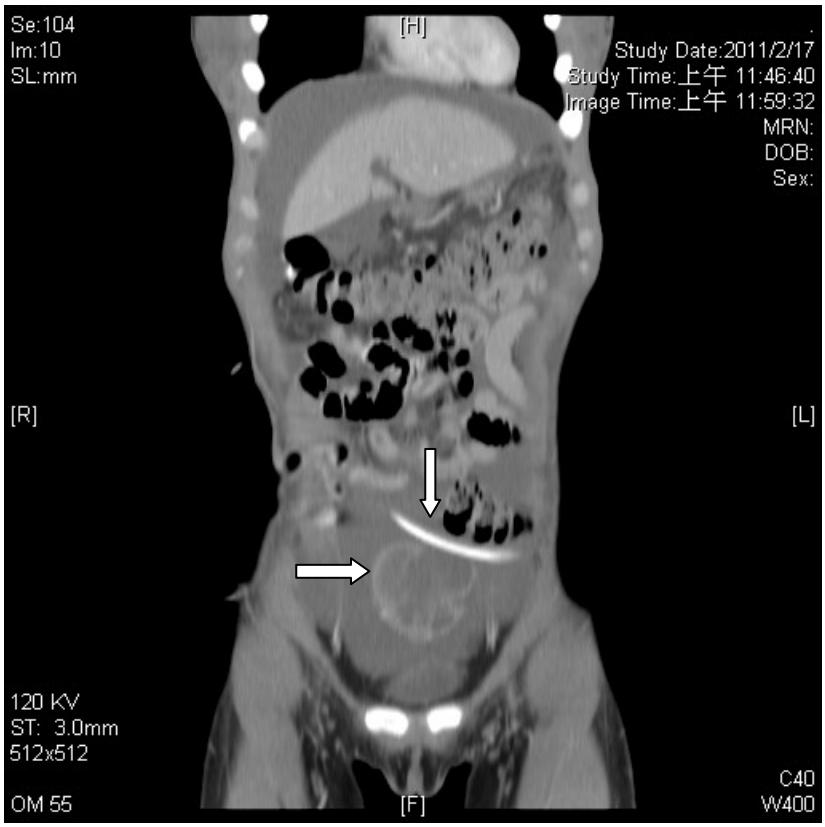
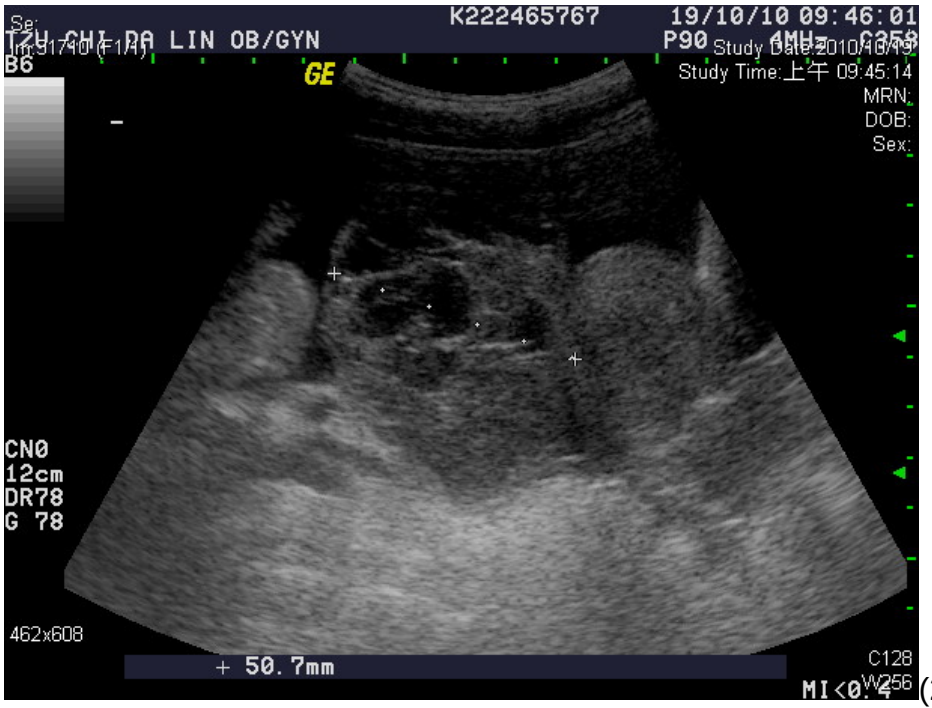
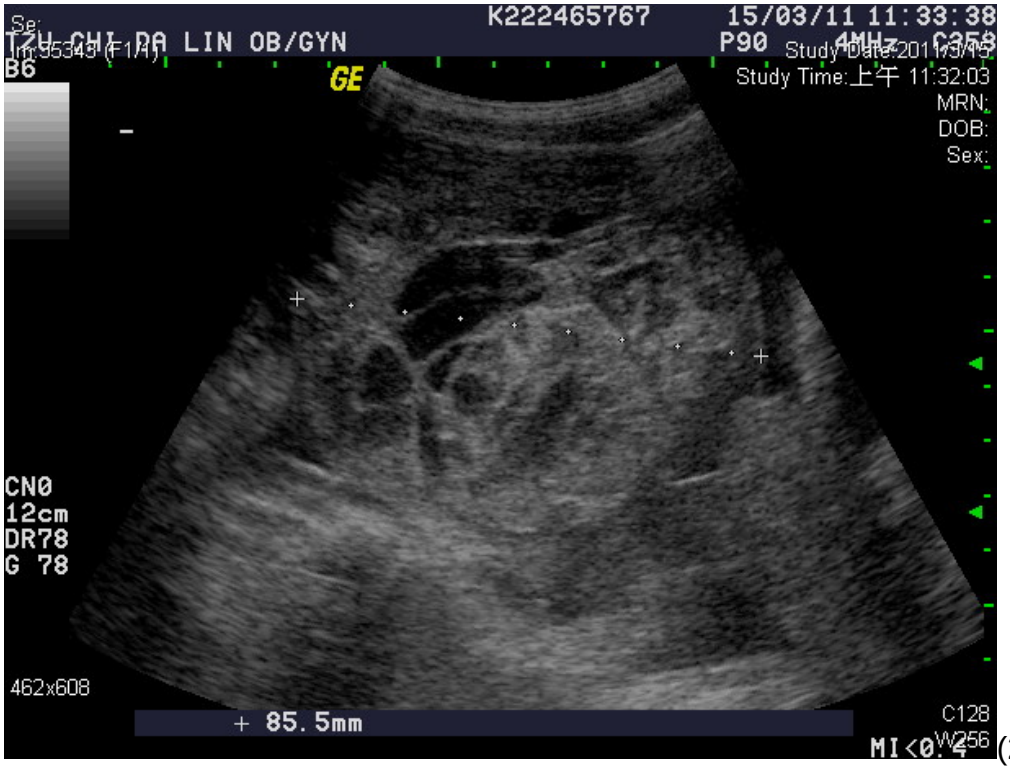


Figure 2. Transabdominal sonography: initial (2a) and 6 months later (2b).



(2a)



(2b)

Figure 3: Pathology of an adult germ cell tumor in this patient.