

Case Report

Pregnancy luteoma followed with massive ascites and elevated CA125 after ovulation induction therapy: a case report and review of literatures

Ying Wang¹, Feng Zhou², Jia-Le Qin³, Zhi-Da Qian⁴, Li-Li Huang⁴

¹The Family Planning Publicity and Technical Guidance Station, Yiwu City, Zhejiang Province, China; ²Department of Pathology, ³Department of Ultrasound, ⁴Department of Obstetrics and Gynecology, Women's Hospital, School of Medicine, Zhejiang University, Hangzhou, Zhejiang Province, China

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Abstract: Objectives: To report a rare case of ovarian tumor with an unusual presentation; an ovarian pregnancy luteoma with massive ascites and elevated CA125 after ovulation induction therapy. Case presentation: A 26-year-old pregnant woman complained lower abdominal distension. Ultrasound imaging showed a solid tumor in the right adnexa and massive ascites. The blood test showed elevated serum level of CA125 and androgens. The patient underwent the right salpingo-oophorectomy, and then the results of blood test were normal and ascites disappeared. Conclusions: pregnancy luteoma followed with massive ascites and increased CA125 after ovulation induction therapy is a very rare case. It is important to provide appropriate medical/surgical intervention without disturbing the pregnancy iatrogenically or causing unnecessary maternal morbidity.

Keywords: Pregnancy luteoma, massive ascites, ovulation induction therapy

Introduction

Pregnancy luteoma is a rare and benign ovarian neoplasm. It is known to result from the hormonal effects during the pregnancy and some cases spontaneously regress after delivering [1]. The lesion occurs usually in the third and fourth decades of life, with the solid ovarian mass, especially bilateral, elevated serum testosterone levels and symptoms of virilization. Grossly, the tumor presents well-circumscribed, soft and fleshy mass, usually accompanied with foci of hemorrhage. The size is variable from a micrometer to over 20 centimeters. The etiological hypothesis is put forward that the luteinized stromal cells are stimulated and proliferated due to the effect of human chorionic gonadotropin; hence these lesions are associated with high levels of androgen [1-3]. We presented a case of an ovarian pregnancy luteoma with massive ascites and elevated serum CA125 after ovulation induction therapy.

Case presentation

A 26-year-old G0 woman with the history of menstrual disorder and treatment of artificial

cycle got a successful pregnancy after ovulation induction. At 15-week gestational age, this patient complained the lower abdominal distension and pain, nausea, vomiting, and felt short of breath.

Transabdominal ultrasonic imaging showed a vital fetal in the uterine with the normal growth parameters, and a 6×5×5 cm right adnexal mass with abundant color Doppler signals (**Figure 1A**). The large-volume ascites were detected in both abdominal and pelvic cavities (**Figure 1B**). The blood test showed the elevated serum level of androgens (52.05 nmol/l), as well as alpha-fetoprotein (AFP, 43.6 ng/ml) and CA125 (625 U/ml). Thus, this patient was suspected with the ovarian malignant tumor and recommended to undergo the right salpingo-oophorectomy. During the operation, the uterus and left ovary were macroscopically normal, while an enlarged right ovary was observed as a solid mass without abdominal metastasis. The massive ascites were sampled and then identified as exudates due to its appearance and the report of total fluid protein (30 g/L).

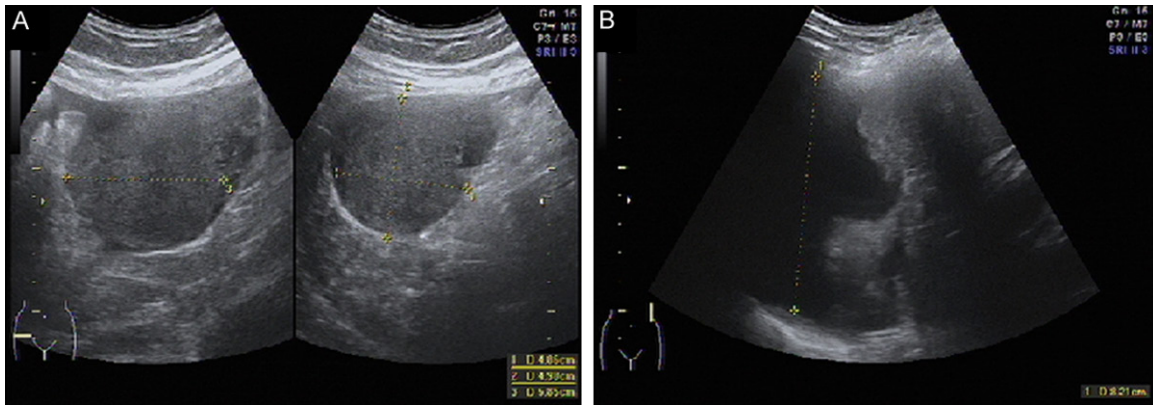


Figure 1. Ultrasound imaging identified a 6×5×5 cm sized right adnexal mass (A). Massive ascites in both abdominal and pelvic cavities (B).

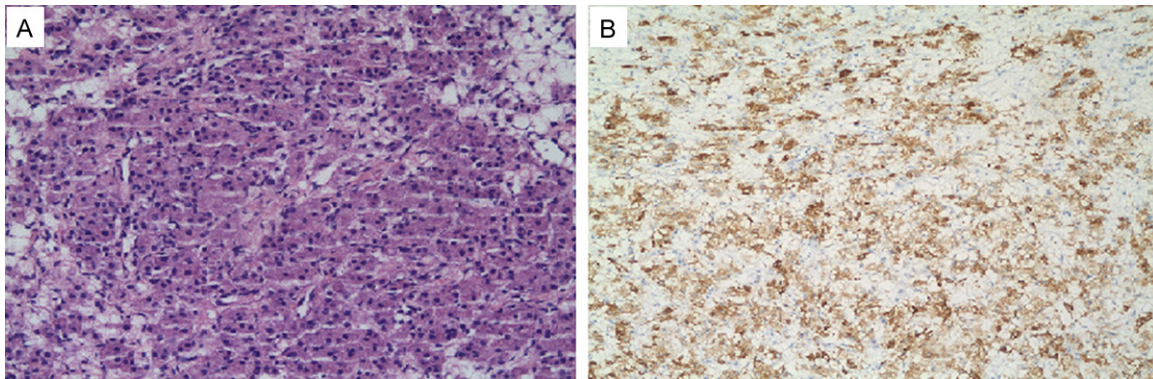


Figure 2. Ovarian nodules were composed of plump, luteinized cells with eosinophilic and often vacuolated cytoplasm, large round to oval vesicular nuclei, and prominent nucleoli (A). Immunohistochemical stains of the right ovarian tumor. Inhibin positively expressed in luteinized cells (B). Magnification: 200×.

Pathologically, the gross character was mainly solid mass embedded with visible oedema, and histopathologic examination showed the cells was abundant eosinophilic foamy cytoplasm, large round or oval vesicular nuclei, and prominent nucleoli (**Figure 2**). Final pathologic examination revealed pregnancy luteoma which is an extremely rare tumor of the ovary.

After the resection, the serum level of tumor markers (CA125, and AFP) and androgen gradually fell down into the recommended ranges. The patient continued pregnancy and underwent cesarean section at a gestational age of 37 weeks due to marginal placental previa. A female infant was born and identified as normal by pediatrician.

Discussion

Luteoma of pregnancy is a benign hyperplastic tumor-like lesion of ovary that was first

described by Sternberg in 1963 [4]. Most of the patients with pregnancy luteoma are asymptomatic and the enlarged ovary has been discovered incidentally until the cesarean section or postpartum tubal ligation. To date, less than 200 cases have been reported.

Approximately 25% of women with pregnancy luteoma have hypersecretion of androgen. Among them, 10~50% mothers present clinical signs of hyperandrogenism, and 60~70% female infants exhibit some degree of virilization [5]. In this case, neither the mother nor the female newborn presented masculine features despite the elevated level of androgen in maternal blood. The potential reason is that the infants are protected by the aromatization of excess maternal androgen to estrogen by the placenta and the high estrogen content of fetal blood [6]. Another reason might be that the tumor was detected before the hormonal sys-

tem of both mother and newborn were affected.

Notably, the massive ascites and an elevated CA125 level in this case resembled malignant tumors. These phenomena have been rarely reported in pregnancy luteomas cases. The overexpression of CA125 could be induced by mechanical stimulation of the mesothelium [7]. Massive ascites during pregnancy is rare and there are few reported cases of recurrence in a subsequent pregnancy. Ovarian hyperstimulation syndrome (OHSS) is a well-known complication of assisted reproductive techniques and is characterized by enlargement of the ovaries and fluid shift from the intravascular compartment to the third space [8]. In this case, there's a history of ovulation induction, which gives rise to the possibility that OHSS might actually be a factor in causing massive ascites. But ascites caused by OHSS is transudate [9] which is not the characteristics of the percolate of this case.

Conclusions

The management of pregnancy luteoma depends upon the symptoms and personal conditions. The asymptomatic pregnancy luteoma that is identified in the third trimester is advised to intensively follow up by imaging (ultrasound or MRI) and expect to regress by itself. On the contrary, the case with atypical presentation or with complications is recommended to undergo surgical intervention for diagnostic or therapeutic purposes. In practice, the surgical invention is to some extent the first choice to treat the patients with severe symptoms and gestational age under the early second trimester.

Disclosure of conflict of interest

None.

Address correspondence to: Dr. Feng Zhou, Women's Hospital, School of Medicine, Zhejiang University, Xueshi Road, Hangzhou 310006, Zhejiang Province, China. Tel: 0086-571-87061501-2022; Fax: 0086-571-87061878; E-mail: 20618235@163.com

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