

A case of massive ovarian edema: ovarian wedge resection by laparoendoscopic single-site surgery (LESS) resulted in complete remission of hyperandrogenic symptoms

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Summary

Massive ovarian edema is a rare entity and easily mistaken as a malignant neoplasm of ovary. This pseudo-tumor occurs predominantly in young women and its presentation varies from being asymptomatic and to being a cause of acute abdomen, such as ovarian torsion. Polycystic ovary syndrome (PCOS) is characterized by elevated androgen and its pathogenesis is associated with endocrine dysfunction. Here the authors present a young woman who showed typical symptoms and signs of PCOS that was finally diagnosed to have massive ovarian edema by laparoendoscopic single-site surgery (LESS) and disappearance of the symptoms of PCOS after resection of edematous pseudo-tumor. This correlation is thought to be the removal of ovarian edema including large proportion of theca tissue which is responsible for overproduction of androgen in ovarian follicles. A 23-year-old woman, a registered nurse at this facility, visited the gynecologic department complaining of six-month amenorrhea and male pattern pubic hair growth. Initial transvaginal ultrasonography showed polycystic morphology in the left ovary and a large right adnexal solid mass with positive Doppler flows. Magnetic resonance imaging was performed and it revealed enlargement of right ovary measuring 9.3 centimeters in largest diameter, with peripheral displacement of follicles and massive ovarian edema was assumed to be a diagnosis. Operation for confirming diagnosis was decided after discussion with patient, as she was anxious for the uncertainty of the nature of the mass. Frozen section pathology report of right ovarian biopsy suggested cystic follicles with edematous stroma. Operation was ended after resection of the two-thirds of the right ovary that showed marked edematous change, while preserving one-third for future fertility. Final pathology confirmed the frozen biopsy. Her symptoms of PCOS that included irregular menstruation and male pattern pubic hair growth were completely improved after the surgery at six-month follow-up.

Key words: Laparoscopic single-site surgery; Massive ovarian edema; Polycystic ovarian syndrome; Wedge resection.

Introduction

Massive ovarian edema is a rare entity and easily mistaken as malignant neoplasm of ovary. This pseudo-tumor occurs predominantly in young women and its presentation varies from being asymptomatic to being a cause of acute abdomen, such as ovarian torsion [1, 2]. Kalston *et al.* first described this pseudo-tumor using the term “massive ovarian edema” in 1969 [3]. According to the World Health Organization’s definition, massive ovarian edema refers to an accumulation of edema fluid within the ovarian stroma separating normal follicular structures [4]. The etiology is not known but assumed to be non-neoplastic, and interference with the venous and lymphatic flow due to torsion of the mesovarium with normal arterial blood flow is thought to be associated with its pathogenesis [5]. The condition is easily misdiagnosed with ovarian malignancy and histologic examination is required to confirm the diagnosis. On the other hand, polycystic ovary syndrome (PCOS) is char-

acterized by elevated androgen in women and its pathogenesis is associated with endocrine dysfunction. Surgical treatment of PCOS including ovarian wedge resection or electrocautery was once considered a treatment option for women pursuing pregnancy. A meta-analysis reported laparoscopic ovarian drilling is as effective as follicle-stimulating hormone treatment in ovulation induction with significantly lower rate of multiple pregnancy. [6] The mechanism is largely unknown, however, it is advised that surgery should be considered as a second-line option for those who are not response to oral agent for ovulation.

Hereby the authors present a case report of a young woman that showed typical symptoms and signs of PCOS with unilateral ovarian enlargement. Her final diagnosis was confirmed to be massive ovarian edema after wedge resection of the affected ovary by laparoendoscopic single-site surgery (LESS), and complete resolution of the symptoms of PCOS occurred after resection of edematous pseudo tumor at six-month follow-up. This correlation is

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Figure 1. — Transvaginal ultrasonography showing a 9×5.5-cm right ovarian mass showing multiple Doppler flows and internal heterogenic foci.

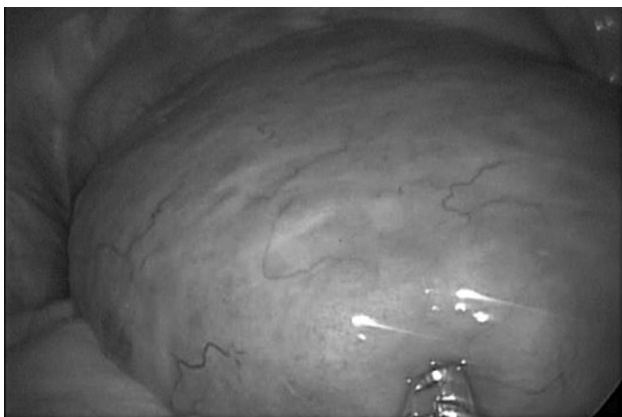


Figure 3. — Gross findings during laparoendoscopic single-site surgery. A diffusely enlarged right ovary is shown.

thought to be the removal of ovarian edema including large proportion of theca tissue, which is responsible for overproduction of androgen in ovarian follicles.

Case Report

A 23-year-old woman, a registered nurse, visited the gynecologic department complaining of six-month amenorrhea and male pattern pubic hair growth. Her past medical history was unremarkable and she attained menarche at the age of 14 years. Initial transvaginal ultrasonography revealed polycystic morphology in left ovary and large right adnexa solid mass with positive Doppler flows (Figure 1). Magnetic resonance imaging was performed. It revealed enlargement of right ovary measuring 9.3 centimeters in largest diameter with peripheral displacement of follicles, and massive ovarian edema was assumed to be a diagnosis (Figure 2). Tumor markers including cancer antigen (CA) 125 and CA 19-9, were below normal reference range.

After discussion regarding her right adnexal mass, she became



Figure 2. — Pelvic magnetic resonance imaging showing peripherally displaced multiple follicles in right ovary with internal high signal intensity in T-2 weighted image. Multiple follicles within the left ovary are shown.

quite anxious due to the uncertainty of the nature of the mass. Considering low malignant potential of the tumor and need for future fertility, she underwent LESS rather than laparotomy. After preoperative discussion, an agreement was reached that the authors would not perform unilateral salpingo-oophorectomy unless the frozen section biopsy revealed malignancy. Gross finding showed intact pedicle of right adnexa and edematous change of the right ovary (Figure 3) Frozen section pathology report suggested cystic follicles with edematous stroma. Operation was ended after resection of the two-thirds of right ovary that showed marked edematous change, while preserving the one-third for future fertility. Final pathology confirmed the frozen biopsy (Figures 4A and B). The patient was discharged uneventfully on postoperative day 3. Her symptoms of PCOS that included irregular menstruation and male pattern pubic hair growth were completely improved after the surgery at six-month follow-up.

Discussion

Massive ovarian edema is a characterized by stromal enlargement of the ovary that might be misdiagnosed as an ovarian malignant tumor leading to radical treatment in young patients [2.] It is often accompanied by masculinization associated with low serum level of gonadotropins caused by overproduction of androgens. With massive ovarian edema, hormonal disturbance which predisposes androgen-dominant microenvironment in ovarian follicles are thought to be related with stromal luteinization and local paracrine factors [7, 8]. Elevated androgens also play a pivotal role in pathogenesis of PCOS. PCOS is the most common endocrine disorder among women in reproductive age [9] and its pathogenesis is currently understood to be associated with disordered ovarian steroidogenesis rather than pituitary gonadotropin secretion [10, 11].

In the present case, a women who is not obese (her body mass index was 22.54), presented with typical symptoms of PCOS including menstrual irregularity and male pattern

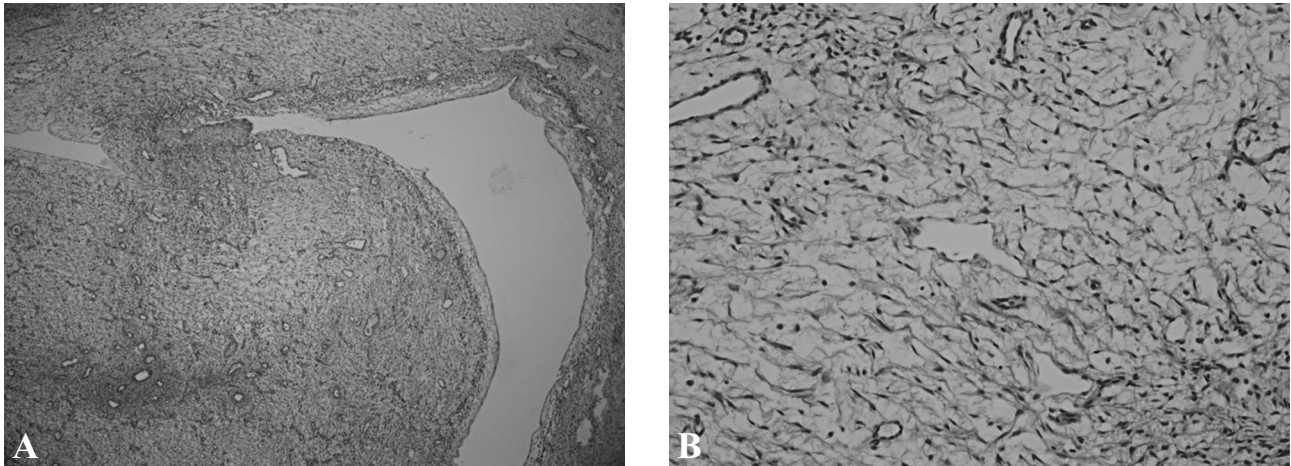


Figure 4. — Microscopic findings of specimen (right ovarian wedge resection). A) H&E staining ($\times 40$) shows cystic follicles with edematous stroma ovary. B) H&E staining ($\times 200$)

pubic hair growth, showing morphologically polycystic ovaries by transvaginal ultrasonography. Correction of symptoms of hyperandrogenism after wedge resection of edematous ovarian stroma may give indication that ovarian stroma might play some crucial roles in the development of androgen-dominance in some PCOS patients. This correlation is thought to be removal of ovarian edema, including a large proportion of theca tissue which is responsible for overproduction of androgen in ovarian follicles. With regards to the treatment of massive ovarian edema, considering that this pseudo-tumor is prevalent in young patient population who required future fertility, wedge resection based on the evidence of frozen section biopsy by minimally invasive laparoscopy rather than unilateral salpingo-oophorectomy by laparotomy seems to be appropriate. Proper preoperative work up for the malignancy potential of the tumor, including serum markers and magnetic resonance imaging in the pelvis, should be considered. Ultrasonography alone may hinder to predict its benign nature since the solid appearance and positive Doppler flows in edematous ovary are indicative of malignant ovarian mass [12].

Physician should keep in mind that ovary-preserving wedge resection by laparoscopy may bring complete resolution of symptoms and pathologic confirmation of ovarian pseudo-tumor in patients complaining of hyperandrogenic symptoms with enlarged ovaries.

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