

# Laparoscopic Plication of Partially Twisted Ovary with Massive Ovarian Edema

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Massive ovarian edema (MOE) is a rare entity characterized by an accumulation of stromal edema fluid and occurs primarily in young women. The etiology is not clear, but is suspected to be the result of partial torsion of the ovary. After the establishment of a correct diagnosis, organ-sparing surgical treatment is the standard treatment. With the assistance of laparoscopy, we diagnosed and managed MOE in a 26-year-old woman who had a 4-year history of primary infertility and intermittent lower abdominal pain that had lasted for more than 6 months. With de-torsion, wedge resection, and plication of the ovary, the patient was successfully relieved of the abdominal pain and experienced no recurrence in the follow-up period. A later spontaneous pregnancy demonstrated the practicality of this conservative treatment. [*J Chin Med Assoc* 2006;69(5):236–239]

**Key Words:** laparoscopic plication, massive ovarian edema, ovarian torsion, polycystic ovary syndrome

## Introduction

Massive ovarian edema (MOE) is a rare entity characterized by an accumulation of edema fluid within the ovarian stroma. It is defined as “marked enlargement of one or both ovaries by an accumulation of edema fluid in the stroma separating the normal follicular structures.”<sup>1</sup> MOE occurs predominantly in young women. The etiology is not clear, but is thought most likely to relate to intermittent or partial torsion of the ovarian pedicle.<sup>2–4</sup> Half of the cases show evidence of torsion at surgery,<sup>4</sup> as in the above-mentioned case. Torsion results in venous and lymphatic obstruction, but not arterial occlusion, so there is no hemorrhage or infarction.<sup>3</sup> The condition occurs frequently on the right side, probably because of the higher right ovarian vein pressure caused by the distinct anatomy of the venous drainage from the right ovary. Lymphedema leads to proliferation of stromal cells and, in some, conversion to lutein cells, resulting in virilization in some patients. Alternatively, primary stromal

proliferation or stromal hyperthecosis can occur, with resultant ovarian enlargement, and subsequent torsion and edema.<sup>3</sup> Therefore, MOE may be classified as primary or secondary, depending on whether or not there is a concomitant pathologic finding predisposing to partial torsion of the mesovarium.<sup>5</sup> Although the etiology of MOEs may be related to torsion, many MOEs without torsion have subsequently been reported.<sup>6–8</sup> After the establishment of a correct diagnosis, organ-sparing surgery is the standard treatment. The first report of MOE was by Kalstone et al in 1969.<sup>9</sup> Although more than 90 cases have been reported worldwide,<sup>2</sup> only 3 cases were treated by laparoscopy.<sup>1,10</sup> None of them was performed with laparoscopic plication of the ovary after wedge resection was performed. With the significant current advantages of laparoscopy in diagnosing and managing benign ovarian diseases,<sup>11–14</sup> we chose this method to accurately diagnose this case of MOE, which was subsequently treated successfully with de-torsion, wedge resection, and plication of the ovary by laparoscopy.

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## Case Report

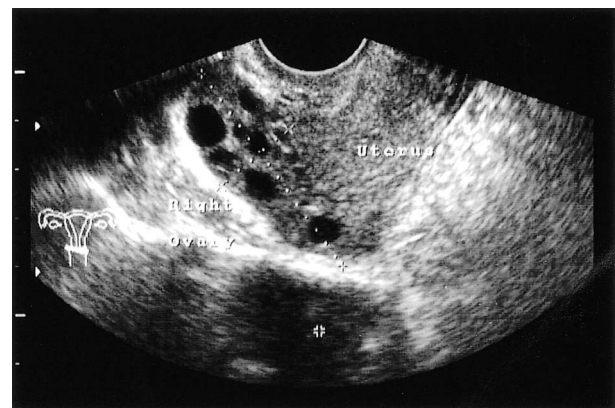
A 26-year-old woman, with a 4-year history of primary infertility, experienced lower abdominal pain that had lasted for more than 6 months. The pain was characterized as being dull without radiation. Her past medical, surgical, and family histories were relatively unremarkable. For the past year, she had suffered from many menstrual problems, including dysmenorrhea, metrorrhagia, and irregular duration. She had also missed her menstruation for 4 months. Physical examination found no hirsutism or virilization. Her abdomen was soft and nondistended. There was moderate tenderness, but no rebound or guarding in the right lower quadrant. Bimanual pelvic examination disclosed a round, tender, slightly mobile mass of right adnexa.

Further examinations revealed a negative Pap smear and a negative urine  $\beta$ -hCG (human chorionic gonadotropin) test. Pelvic ultrasound demonstrated a septated 4 × 3-cm mass with 2 cystic components in the left ovary, a 5 × 2-cm left ovary with numerous follicles, and a normal-appearing uterus (Figure 1). A tumor flow was not performed, partly because malignancy was unlikely, and, of most importance, colored Doppler ultrasound was unavailable. A hormone profile (the blood was sampled on day 3 during the menstruation) showed a marked elevation of serum luteinizing hormone (LH) level (11.8 mIU/mL), but others were all within normal limits, including: follicle-stimulating hormone (FSH) (3.95 mIU/mL); testosterone (0.3 ng/mL); 17  $\alpha$ -hydroxyprogesterone (0.7 ng/mL); dehydroepiandrosterone (DHEA); insulin (18.5 ng/mL); DHEA sulfate (5.4  $\mu$ mol/L); prolactin (12.4  $\mu$ g/L); and thyroid function (1.4 ng/dL of free T4 and 1.1  $\mu$ U/mL of thyroid-stimulating hormone). Complete blood count, electrolytes, fasting sugar, CA125, liver, and renal function tests were within normal values.

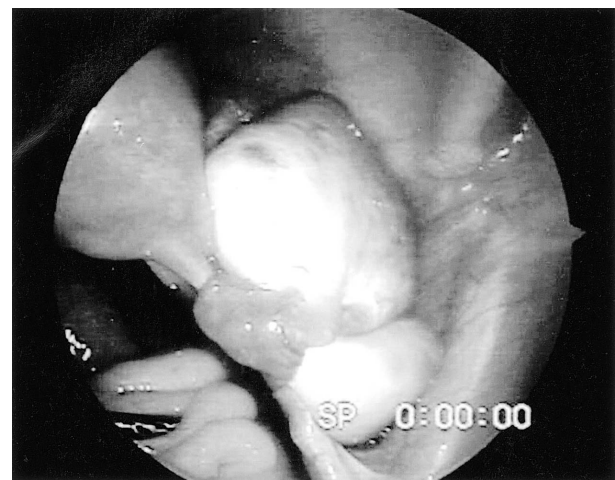
Diagnostic laparoscopic surgery was performed for the following reasons: more than 6 months of lower abdominal pain, primary infertility, and clinical suspicion of polycystic ovary syndrome (PCOS). The right meso-ovarium and the ipsilateral fallopian tube were twisted counter-clockwise (Figure 2). The involved fimbria was edematous, but with no ischemia. The right ovary was enlarged and edematous (5 cm), and was rotated and displaced in the anterior aspect of the broad ligament. The left ovary was slightly enlarged (4 cm). The uterus appeared normal in shape and size. After untwisting the involved ovarian ligament and the fallopian tube, a wedge resection of the bilateral

ovarian cysts was performed. The right ovary was plicated by fixing the utero-ovarian ligament to the posterior uterine wall. A cut section of right ovarian tissue was grayish yellow, with a scant amount of bloody fluid. A small piece of left ovary was removed. Frozen biopsy was not done because malignancy was unlikely in the gross view. Final microscopic examination showed stromal edema with a rim of normal ovarian tissue in the bilateral ovaries. Figures 3A and B demonstrate typical findings of massive ovarian edema from the right ovary.

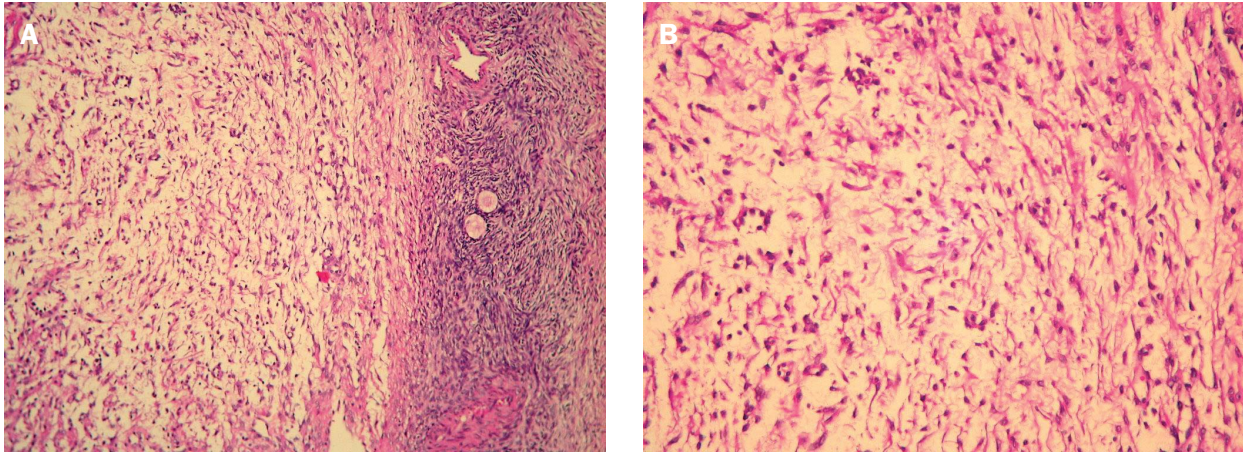
The final diagnosis was right-sided MOE (by pathology) and PCOS (by clinical findings)<sup>15</sup> with partial torsion (by operative finding). The patient has been symptom-free for more than 30 months. During follow-up, she had regular menstruation and became pregnant without the assistance of any artificial reproductive techniques. She later delivered a normal baby.



**Figure 1.** Transvaginal ultrasound shows an enlarged ovary containing numerous different-size follicles and a normal-appearing uterus.



**Figure 2.** Laparoscopy demonstrates that the right meso-ovarium and the ipsilateral fallopian tube were twisted counter-clockwise.



**Figure 3.** (A) Left side of the ovary shows edematous change of the stroma with low cellularity and right side demonstrates normal ovarian tissue (dense cellular area, blue color with 2 primitive follicles in the right center) (hematoxylin and eosin,  $\times 100$ ). (B) High-power field shows marked edematous change with low cellularity (hematoxylin and eosin,  $\times 200$ ).

## Discussion

MOE is usually found in young women; however, premenarcheal<sup>16</sup> or menopausal<sup>17</sup> females have been reported. The initial presentation might be pelvic pain, menstrual abnormalities, infertility, early puberty, or virilization. Acute pain can occur and, presumably, is secondary to torsion.<sup>3</sup> Menstrual irregularity is present in most patients, and subsides after treatment.<sup>4</sup> Virilization can be present in about 25% of cases.<sup>3</sup> The incidence of MOE is unclear, because the pathologic diagnosis of MOE is unfamiliar to the majority of pathologists, and because the diagnosis of MOE has always been retrospective. After reviewing our hospital-based data of adnexal torsion, which was managed by surgery through 1996, only this case was reported as MOE. The incidence was 1 in 79 cases of adnexal torsion managed by surgery. Their final pathology included mature cystic teratoma (44 cases), corpus luteum (23 cases), mucinous cystadenoma (6 cases), and others (5 cases), including MOE (1 case).

The preoperative diagnosis of MOE is relatively difficult, partly because the diagnosis has always been retrospective—only surgery could identify it correctly. Recently, there have been several reports describing the preclusion of MOE aided by ultrasound or magnetic resonance.<sup>4,6</sup> Multiple ovarian follicles located at the peripheral cortex of an enlarged ovary are suggested to be a diagnostic indicator of MOE,<sup>6</sup> although this ultrasound feature is also a diagnostic criterion for typical PCOS.<sup>18</sup> The criteria fulfilling sufficient specificity and sensitivity to define the PCO should have at least 1 of the following: either 12 or more follicles measuring 2–9 mm in diameter, or increased

ovarian volume ( $> 10 \text{ cm}^3$ ).<sup>18</sup> Microscopically, diffuse edema was detected in the stroma, which is consistent with a diagnosis of MOE. However, as for the size of the affected ovary, “massive” may be a misnomer, because most patients have presented with minimal ovarian enlargement. By contrast, there was no doubt in the diagnosis of PCOS in this case, since the ovarian volume was significantly increased ( $> 10 \text{ cm}^3$ ). Therefore, a more precise diagnosis might be made with a combination of PCOS and MOE. That is why the fertility outcome of this patient was excellent, since we performed the wedge resection of the ovary to improve the hormonal imbalance in PCOS.

Oophorectomy or salpingo-oophorectomy previously was the chosen procedure, partly because of fear of recurrence. Sometimes, either benign or malignant ovarian tumors should be made as a differential diagnosis.<sup>19</sup> Metastatic disease of another malignancy may be a cause of MOE if the ovarian lymphatic vessel is obstructed by carcinoma cells.<sup>7,8</sup> Because resolution of the affected ovaries can be expected after wedge resection, a wedge resection with or without a subsequent frozen section to preserve the ovarian function is mandatory for younger patients.<sup>19</sup> Complete preoperative evaluation, careful inspection for ovary and other organs or tissues in the abdominal cavity, and optional performance of frozen biopsy during surgery could prevent unnecessary oophorectomy, especially in young women with MOE. Conservative treatment is currently the preferred principle of management. To avoid repeated torsion, ovarian suspension with a fixing of the ovary to the uterus or broad ligament is recommended.<sup>20</sup> Laparoscopic surgery is less aggressive and less



traumatic in comparison with laparotomy. For the conservative treatment of MOE, laparoscopy should be considered. By modifying the laparoscopic procedure of Kocak et al,<sup>1</sup> who performed wedge resection to manage MOE by laparoscopy, we describe the first case of MOE treated with deep wedge resection. This was done to improve hormone balance. Plication of the ovary was for decreasing the possibility of further torsion of the ovary. The lack of recurrence and spontaneous pregnancy in this patient has demonstrated the practicality of this conservative treatment.

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