

Massive ovarian edema with paraovarian cyst torsion treated with laparoscopic surgery: A case report

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Abstract. Massive ovarian edema is a rare gynecological entity resembling a solid ovarian tumor due to the accumulation of edematous fluid within the ovarian stroma. This condition can be easily mistaken for a neoplasm, resulting in overtreatment by removal of the whole affected ovary. The present study describes the case of a 28-year-old woman who experienced massive ovarian edema with paraovarian cyst torsion treated with laparoscopic surgery. The patient experienced lower abdominal pain lasting for 1 week and visited a local clinic. The ultrasonographic examination revealed two loculated ovarian masses and the patient was then referred to the hospital. Transvaginal ultrasonographic examination revealed a 77.9-mm cystic lesion and a 57.7-mm solid lesion in the left adnexa. A magnetic resonance imaging examination revealed a 55-mm lesion with multiple peripheral ovarian follicles, which was isointense on T1-weighted images and hyperintense on T2-weighted images, and a 75-mm cystic lesion, without a solid component, which was hypointense on T1-weighted images and hyperintense on T2-weighted images in the left adnexa. There were no observed abnormalities of the right adnexa or uterus. Laparoscopic surgery was performed, based on a clinical suspicion of massive ovarian edema with paraovarian cyst torsion. Intraoperatively, a paraovarian cyst was identified in the left adnexa that was twisted 360°. The size of the enlarged left ovary was reduced to almost normal following the detorsion of the left adnexa. The final diagnosis was that of a massive ovarian edema, which was treated by resecting the paraovarian cyst, while preserving the whole left ovary. The pathological examination of the resected paraovarian cyst revealed a serous cystadenoma. Therefore, the present study suggests that the presence of massive ovarian edema should be taken into consideration when encountering

a complex solid ovarian mass with multiple peripheral ovarian follicles, particularly in cases with a history of recurrent abdominal pain.

Introduction

Massive ovarian edema is a rare gynecological entity resembling a solid ovarian tumor due to the accumulation of edematous fluid within the ovarian stroma, first described by Kalstone *et al* in 1969 (1) and defined by the World Health Organization as an accumulation of edematous fluid within the ovarian stroma separating normal follicular structures. Massive edema of the ovary is a rare condition affecting mainly young women (mean age, 20 years); however, premenarcheal (2) or menopausal women (3) have been reported to be affected as well. Massive ovarian edema is characterized by a build-up of interstitial fluid without neoplastic changes, and it is mainly considered to be the consequence of torsion of the ovary, despite the fact that there are several reported cases without torsion observed during surgery (4-7). However, this condition can be easily mistaken for a neoplasm, resulting in overtreatment by the removal of the whole affected ovary. Conservative treatment is recommended for this entity, particularly when preservation of fertility is important. Therefore, the accuracy of preoperative diagnosis should be improved by taking massive ovarian edema into consideration when solid enlargement of the ovary is detected.

The present study describes the case of a 28-year-old woman who presented with massive ovarian edema with paraovarian cyst torsion and was successfully treated with conservative laparoscopic surgery owing to the accurate preoperative diagnosis.

Case report

A 28-year-old woman (gravida 1, para 1) visited a local clinic due to mild left lower abdominal pain lasting for 1 week. The patient's medical history and family history did not reveal any abnormalities. Her menstrual cycle was regular at 28 days, and she had experienced menarche at the age of 12 years. An ultrasonographic examination revealed two loculated left ovarian masses and the patient was referred to the Osaka City University Hospital (Osaka, Japan) for further evaluation.

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The patient did not suffer from abdominal pain when she visited the hospital. Upon a physical examination, the uterus was found to be anteverted, anteflexed and resembled a hen's egg in size; a slightly painful left adnexal mass was palpated, whereas the right adnexa was not palpable during a bimanual examination. There was no hirsutism or signs of virilization. The transvaginal ultrasonographic examination performed at the hospital revealed a 77.9-mm cystic lesion and a 57.7-mm solid lesion with multiple peripheral ovarian follicles in the left adnexa (Fig. 1). The magnetic resonance imaging (MRI) examination revealed a 55-mm lesion with multiple peripheral ovarian follicles, which was isointense on T1-weighted images and hyperintense on T2-weighted images, and a 75-mm cystic lesion without a solid component, which was hypointense on T1-weighted images and hyperintense on T2-weighted images in the left adnexa (Fig. 2). The right ovary was normal, exhibiting only a small functional cyst. There were no observed abnormalities of the right adnexa or the uterus. A laboratory examination revealed normal complete blood count, electrolyte levels, liver and renal function tests, C-reactive protein and lactate dehydrogenase levels; the serum level of carbohydrate antigen (CA) 125 was 13 U/ml (normal limits, ≤ 35 U/ml) (8), while that of CA19-9 was < 2 U/ml (normal limits, ≤ 37 U/ml) (9), that of carcinoembryonic antigen was 1.3 ng/ml (normal limits, < 5 ng/ml) (10) and that of Sialyl-Tn was 20.6 U/ml (normal limits, ≤ 45 U/ml) (11), which were all within normal limits.

Laparoscopic surgery was performed based on the clinical suspicion of a massive ovarian edema with paraovarian cyst torsion. A paraovarian cyst was identified in the left adnexa; the left adnexa was twisted 360° , and the left ovary appeared enlarged, with a white and opaque external surface (Fig. 3A). The right adnexa and uterus were normal in appearance and there was no ascites. Following detorsion of the left adnexa, the size of the enlarged left ovary gradually returned to almost normal (Fig. 3B). The diagnosis of a massive ovarian edema was considered likely, due to the shrinkage in size following detorsion and the findings of the pre-operative ultrasonographic and MRI examinations. A substantial amount of paraovarian cyst fluid was drained using the SAND balloon catheter (Hakko Shoji Co., Ltd.), as previously described by Yagihashi *et al* (12) without spillage of the cyst contents; the paraovarian cyst was resected, resulting in the successful preservation of the whole left ovary. Following surgery, the pathological examination using hematoxylin and eosin staining (performed by the Department of Pathology, Osaka City University Graduate School of Medicine, Osaka, Japan) as routine clinical work of the paraovarian cyst revealed a serous cystadenoma without any malignant characteristics (Fig. 4). At the 1-month post-operative follow-up visit at the hospital, an ultrasonographic examination confirmed normal-sized bilateral ovaries and uterus (Fig. 5).

In the case presented herein, the diagnosis of massive ovarian edema was not confirmed by a pathological examination; however, the diagnosis of a massive ovarian edema was based on the other available evidence, including the identification of multiple ovarian follicles at the periphery of the enlarged ovary, which shrank following detorsion.

Discussion

Massive ovarian edema was first described in 1969 by Kalstone *et al* (1) as a 'massive, solid enlargement of the ovary associated with interstitial edema, without neoplastic changes'. The etiology is unknown; however, it may result from the partial intermittent torsion of the ovary causing venous and lymphatic obstruction, to the extent that it interferes with venous and lymphatic drainage, but is not sufficient to cause necrosis. The removal of high-molecular-weight proteins from the interstitium of the ovary can be hindered by any degree of compression at the ovarian hilum, which is followed by an increase in the osmotic pressure, resulting in edema formation. This causes the shutdown of the lymphatic capillary pores and results in a vicious cycle of accumulation of increased amounts of fluid in the interstitium of the ovary. According to this mechanism, lymphatic blockage may occur even in the absence of complete torsion (13). Massive ovarian edema is a rare entity affecting mainly young women. Therefore, it is crucial to recognize this entity, as it is often misdiagnosed as a malignant ovarian tumor, placing the young patient at risk of overtreatment and resulting in the loss of hormonal function and fertility. The most common presenting symptoms include recurrent intermittent abdominal pain, abdominal distension or an abdominal mass, menstrual abnormalities, infertility and, in some cases, virilization and precocious puberty (14). The excess amounts of androgen secreted by luteinized stromal cells in the edematous ovary are considered to cause androgenic manifestations (4). In cases in which the torsion is acute, the most common symptom is acute abdominal pain that may mimic an acute abdomen. In the cases in which the torsion is gradual, a provoked stromal luteinization may cause virilization in the patient (15).

There are two types of massive ovarian edema, depending on the presence or absence of concomitant pathological findings predisposing to the partial torsion of the mesovarium. Primary massive ovarian edema is an entity without a concomitant pathology, whereas secondary massive ovarian edema is an entity superimposed on already diseased ovaries (16). The most favored hypothesis for the etiology is obstructed venous and lymphatic circulation, but not arterial blood flow, due to the complete or partial torsion of the ovary, leading to the development of a massive ovarian edema (13). As a result, stromal cell luteinization is induced, due to the response of the edematous ovary to torsion and subsequent ischemia, which causes hormone production (17). There are estrogen and progesterone receptors in stromal cells, and mechanical stimulation due to stretching of the stroma by the edematous fluid may lead to hormone-related symptoms (13,14,18). Another explanation for the hormone-related changes is the deregulated expression of a local paracrine factor, such as epidermal growth factor, insulin-like growth factor or cytokines (18). Secondary massive ovarian edema occurs in diseased ovaries, such as those with an ovarian mass or cyst-like ovarian capillary hemangioma (19), serous and mucinous cyst adenomas (6), mature cystic teratoma (20), ovarian fibrothecoma (21), polycystic ovary syndrome (22) and Meigs syndrome (23), and may also occur as a consequence of drug treatment for ovulation induction (24) and in some

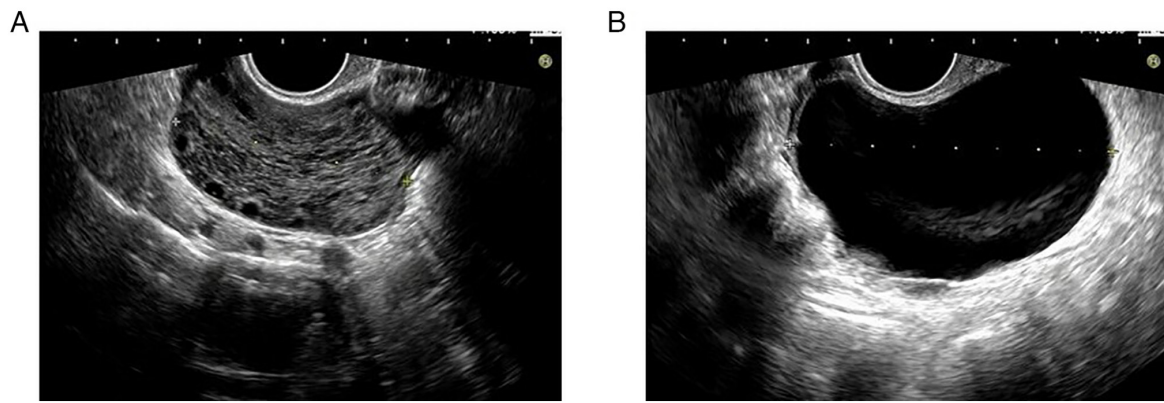


Figure 1. Pre-operative transvaginal ultrasonographic images. (A) A 57.7-mm solid lesion with multiple peripheral ovarian follicles and (B) a 77.9-mm cystic lesion were identified in the left adnexa.

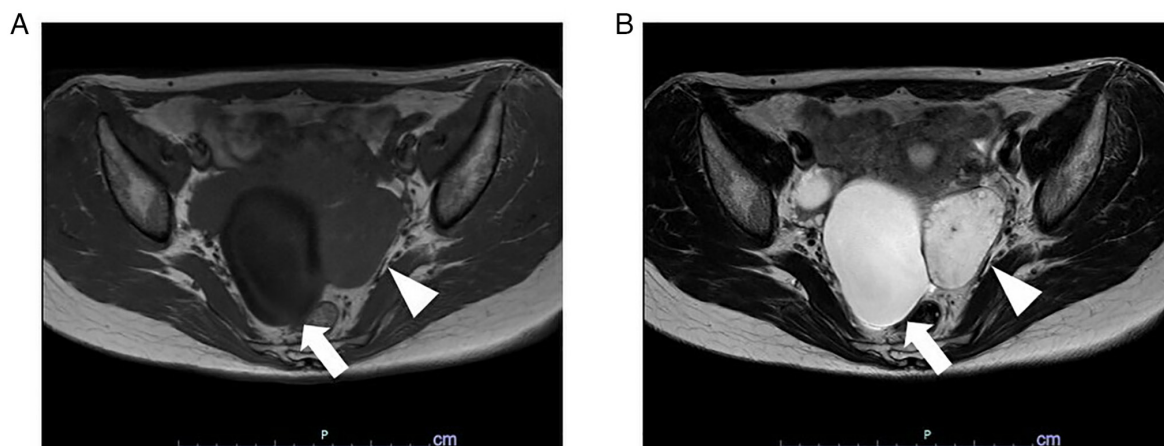


Figure 2. Preoperative MRI examination. (A) T1-weighted and (B) T2-weighted axial images. A 55-mm lesion with multiple peripheral ovarian follicles, which was isointense on T1-weighted images and hyperintense on T2-weighted images, and a 75-mm cystic lesion without a solid component, which was hypointense on T1-weighted images and hyperintense on T2-weighted images, were identified in the left adnexa. The right ovary was normal, exhibiting only a small functional cyst. The white arrows indicate the left paraovarian cyst. The white arrowheads indicate the left ovary.

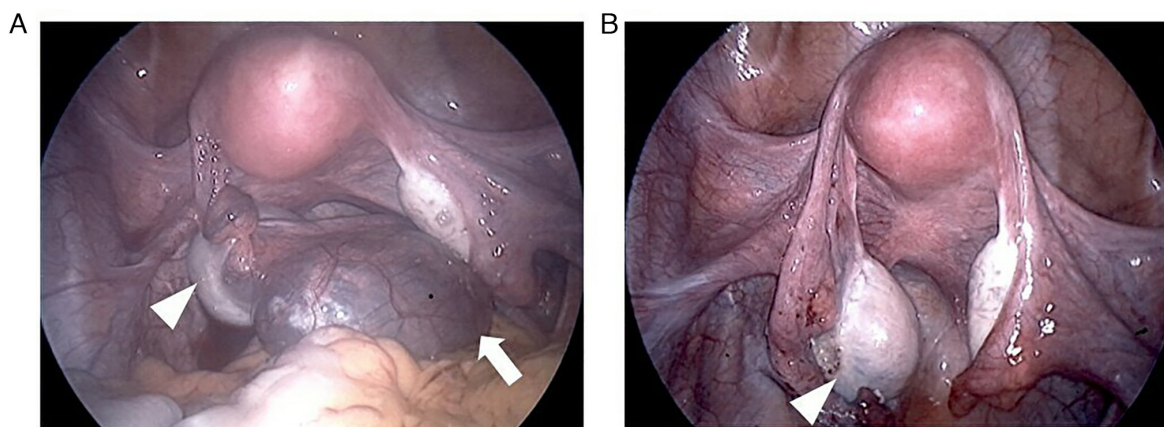


Figure 3. Intraoperative findings during laparoscopic surgery. (A) Prior to detorsion, the left adnexa with the paraovarian cyst was twisted 360° and the left ovary was enlarged, with a white and opaque external surface. The right ovary was normal in size. (B) Following detorsion and the removal of the paraovarian cyst, both ovaries appeared normal in size. The white arrow indicates the left paraovarian cyst. The white arrowheads indicate the left ovary.

malignancies (25,26). Malignancies reported to cause ovarian lymphatic vessel obstruction by metastatic carcinoma cells include gastric carcinoma (26), uterine cervical cancer (25) and lymphangitis carcinomatosa (27).

Although there are no diagnostic imaging criteria, ultrasonography and MRI have been reported to be the most useful modalities for diagnosing this entity. Umesaki *et al* (28) reported that a solid ovarian tumor with

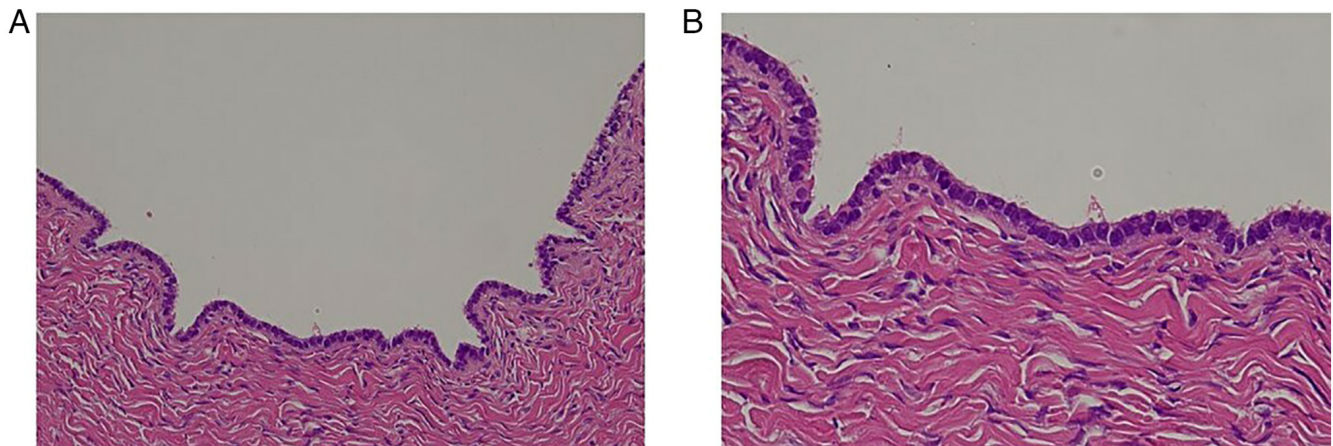


Figure 4. Pathological findings of the resected paraovarian cyst (hematoxylin and eosin staining). The cyst was lined by ciliated columnar epithelium without atypia, indicating serous cystadenoma. Magnification, (A) x20 and (B) x40.

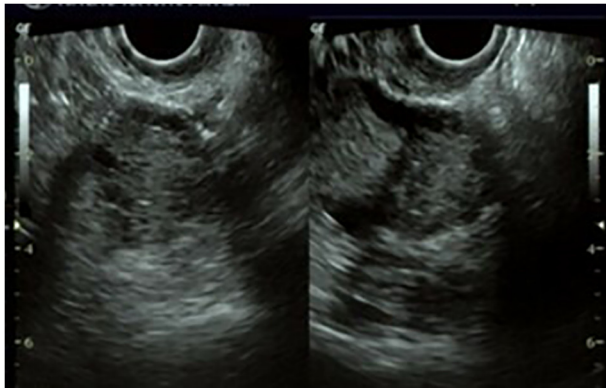


Figure 5. Transvaginal ultrasonography was performed 1 month after surgery. Left panel, right ovary; right panel, left ovary. Both ovaries were normal in size.

multiple peripheral ovarian follicles on ultrasonographic examination may indicate a possible preoperative diagnosis of massive ovarian edema. Furthermore, another report by Umesaki *et al* (29) using MRI demonstrated that the main indicator of massive ovarian edema is an enlarged ovary with edematous stroma exhibiting high intensity on T2-weighted images, with multiple ovarian follicles pressed towards the peripheral cortical area of the ovary by the edematous fluid accumulated within the ovarian stroma. Similarly, Hall *et al* (30) also reported that the presence of multiple ovarian follicles situated around the periphery of the cortex of the enlarged ovary on MRI is a crucial sign that indicates massive ovarian edema. Therefore, the finding of multiple ovarian follicles located at the peripheral cortex of an enlarged ovary is considered to be a crucial diagnostic evidence of massive ovarian edema.

The morphological characteristics of massive ovarian edema are as follows: The external surface of the ovary is usually white and opaque, or grayish and glistening; on cross section the internal surface is grey in color and typically watery fluid oozes out from the cut surface due to the edematous pressure; and, in the majority of cases, the ovary has a rubbery consistency (13,15).

Histologically, the ovarian architecture is preserved, but with an edematous and hypocellular ovarian stroma, and a thickened and fibrotic outer cortex. In cases with endocrine symptoms, a cluster of luteinized stromal cells is occasionally observed (31). There is diffuse edema confined to the medullary stroma and the spared cortex. Small subcortical follicular cysts, and uniformly dilated blood and lymphatic vessels are observed (15). Necrosis and hemorrhage are unusual, as the torsion commonly causes venous and lymphatic obstruction, but not arterial occlusion (31). Focal stromal luteinization has been noted in some of the studied cases, and it is considered to be a mechanical stimulation of proliferation and luteinization of stromal cells induced by stretching of the stroma due to lymphedema (14). Nogales *et al* (15) reported no histopathological evidence of a proliferative process, such as fibromatosis, and concluded that massive edema of the ovary is a reactive, non-proliferative state of specific stromal cells due to torsion of the ovary.

In the case present herein, the MRI findings were typical of a massive ovarian edema, demonstrating an enlarged ovary with edematous stroma that appeared hyperintense on T2-weighted images, with multiple ovarian follicles pressed towards the peripheral cortical area of the ovary. Based on the characteristic MRI images and prior knowledge of this entity, the condition was diagnosed pre-operatively as a massive ovarian edema. The fact that the size of the enlarged ovary returned to almost normal following detorsion of the adnexa intraoperatively further confirmed the suspected clinical diagnosis, and the affected adnexa was preserved by simply removing the paraovarian cyst.

Due to the lack of awareness of this entity and its resemblance to a neoplastic disease, more aggressive treatment than necessary is often undertaken, with a resultant loss of fertility and hormonal function. The differential diagnosis of massive ovarian edema should be taken into consideration in young women presenting with a complex solid ovarian mass with multiple peripheral ovarian follicles, particularly in cases with a history of recurrent abdominal pain. An accurate pre-operative evaluation, with careful inspection of the ovary and other organs or tissues in the abdominal cavity, may prevent unnecessary surgery, including oophorectomy, particularly in young women with massive ovarian edema.

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Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Authors' contributions

TF and TS conceived and designed the study. TF, KI, MY, MK, TI and TY acquired, examined and interpreted the data of the patient. TF and TS drafted and revised the manuscript. TF and TS confirm the authenticity of all the raw data. All the authors have read and approved the final manuscript.

Ethics approval and consent to participate

Written informed consent was obtained from the patient depicted in the present case report.

Patient consent for publication

Written informed consent was obtained from the patient for the publication of the case details and any accompanying images.

Competing interests

The authors declare that they have no competing interests.

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