International Journal of Clinical Obstetrics and Gynaecology

ISSN (P): 2522-6614
ISSN (E): 2522-6622

www.gynaecologyjournal.com
2024; 8(4): 131-133
Received: 08-06-2024
Accepted: 12-07-2024

Dr. Kevin Kiran Rambhia
M.S., Assistant Professor, Department of Obstetrics and Gynaecology, Lokmanya Tilak Municipal Medical College and General Hospital, Mumbai, Maharashtra, India

Dr. Yajnaseni Banerjee
M.S., Assistant Professor, Department of Obstetrics and Gynaecology, Topiwala National Medical College, Mumbai, Maharashtra, India

Corresponding Author:
Dr. Kevin Kiran Rambhia
M.S., Assistant Professor, Department of Obstetrics and Gynaecology, Lokmanya Tilak Municipal Medical College and General Hospital, Mumbai, Maharashtra, India

A rare case of bilateral massive ovarian oedema masquerading as torsion

Dr. Kevin Kiran Rambhia and Dr. Yajnaseni Banerjee

DOI: https://doi.org/10.33545/gynae.2024.v8.i4b.1488

Abstract
Massive ovarian oedema is a rare, poorly understood condition with highly varied clinical presentation. Partial torsion of the vascular pedicle compromising veno-lymphatic drainage without affecting arterial supply has been hypothesized to be the aetiology. Thus far only about 200 cases have been documented in literature, the first one being in 1969. These patients may present with acute abdomen, menstrual irregularities, abdominal masses, abdominal distension or virilization. Despite generally being benign in nature, and lacking any hallmark radiological signs, imaging frequently raises the suspicion of malignancy. In up to 85% cases there is no primary ovarian pathology. Yet, a large majority of cases undergo extensive, often unnecessary extirpative surgery, negatively affecting hormonal function with implications on future reproductive potential. A review of literature suggests that judicious use of ovarian wedge resection with symptomatic management is likely to yield better outcomes.
We present a case of a 19-year-old girl with acute abdomen with imaging suggestive of bilateral ovarian torsion and the sequence of events that unfurled. Our case further emphasizes the lacunae in our understanding of this elusive condition and the need for more research to establish robust, evidence-based management protocols.

Keywords: Benign ovarian oedema, fertility sparing surgery, massive ovarian oedema, ovarian pseudo-tumour, ovarian wedge biopsy, ovarian torsion

Introduction
Massive ovarian oedema is a rare, benign condition characterized by a significant solid enlargement of the ovary, often associated with interstitial oedema but without neoplastic changes [1]. Having been first described in 1969 by Kalstone *et al.*, thus far only about 200 cases have been described in medical literature [2]. It typically affects women aged 6 to 33, with sporadic occurrences in menopausal women, and can occur in one or both ovaries, including during pregnancy [3]. The exact cause is unclear, but it is believed to result from interference with venous and lymphatic flow due to recurrent partial or complete torsion of the mesovarium. In these cases, the arterial blood flow is unimpeded, preventing gangrene or necrosis [4]. Other differentials include retroperitoneal lymphoma, metastatic carcinoma, and polycystic ovary syndrome (PCOS). The clinical presentation depends on how quickly the torsion occurs. Rapid torsion causes sudden abdominal pain, which can resemble appendicitis. Slow torsion can result in stromal luteinization, leading to symptoms like virilization, menstrual irregularities, precocious puberty, or Meigs syndrome [5].
It is essential to recognize this condition because it is frequently misdiagnosed as malignancy, resulting in unnecessary interventions that can harm hormonal function and fertility. Diagnosis relies entirely on histo-pathologic examination, as no clinical or laboratory tests can confirm it.
Case Report:
A 19-year-old unmarried girl presented to the ER with features of acute abdomen (abdominal pain, vomiting, fever). Her vitals were as follows: Pulse=120/min, BP=90/60 mm Hg, afebrile, moderate pallor, features of dehydration. Per abdominal examination showed generalised tenderness without guarding/rigidity or any palpable mass. No obvious features of virilization were noted. Her routine laboratory investigations were suggestive of moderate hypochromic microcytic anaemia (Hb=8g/dl), elevated white cell count (15000/µl) and raised ESR (50 mm/hr). Urine routine microscopy was normal. Preliminary ultrasound suggested moderate hepatomegaly, moderate ascites and bilateral enlarged ovaries (left ovary 80 cc, right ovary 75 cc) with heterogenous echotexture and cystic changes in ovarian stroma.

Ultrasound-guided ascitic fluid tapping was done for routine/microscopy [90% polymorphs, 10% lymphocytes, no malignant cells, TLC 660 cells/ µl], ADA levels [54 IU/L], culture-sensitivity [no growth] and CB-NAAT for tuberculosis [negative for Mycobacterium Tuberculosis]. After initially instituting conservative management comprising of intravenous antibiotics, fluid resuscitation and transfusion of 1 pint packed red cells, the patient was monitored in ICU setting with multidisciplinary involvement (general surgery, internal medicine, intensivist).

In view of worsening clinical condition (onset of abdominal distension, worsening pain scores, rising white cell count-25000/µl, thrombocytopenia 80000/µl, INR-1.4), further imaging was sought and tumour markers were sent [α-fetoprotein, Carcino embryonic antigen, CA-125, β-human chorionic gonadotrophin, CA-19.9 were within normal limits]. Contrast enhanced ultrasound showed delayed time to peak (280 ms) in bilateral ovaries, adnexal doppler showed no colour flow in bilateral ovarian vessels with both ovaries enlarged and placed in midline in pouch of Douglas. This strongly suggested bilateral ovarian torsion with ovarian infarct. (Image 1).

Exploratory laparotomy was performed in conjunction with general surgeons; the findings were as follows: Straw coloured ascitic fluid of approximately 1-1.5 litres which was sent for cytology, bilateral bulky ovaries (approximately 7 x 5 x 4 cm each) with no evidence of ovarian torsion, uterus and bilateral fallopian tubes were grossly unremarkable (Image 2). Wedge biopsies were taken from both ovaries. Bowel tracing was done, omental and peritoneal biopsies taken. Liver, gall bladder, stomach and spleen were grossly examined and showed no evidence of any pathology.

Postoperatively, the patient appeared to be clinically stable for the first 24 hours, was extubated and maintaining oxygen saturation on room air but subsequently suffered recurrent fever spikes with an acute episode of desaturation for which she was re-intubated and mechanical ventilation was started. Investigations on post-operative day 3 were as follows: Hb 8.2 g/dl, TLC 37000/µl, platelet count 88000/µl, INR 1.2, Serum creatinine 1.2 mg/dl, Total Bilirubin 6.9 g/dl, Direct Bilirubin 5.2 g/dl, ALT 24 IU/L, AST 79 IU/L, blood culture no growth, urine culture no growth, 2d echocardiography ejection fraction 60%, no evidence of vegetation/clot, thin rim of pericardial effusion present, mild mitral and tricuspid regurgitation present, HRCT-Suggestive of bilateral pulmonary infiltrates likely due to Acute Respiratory Distress Syndrome (ARDS), Ascitic fluid cytology-consistent with pre-operative ascitic tap.

Following a tumultuous course in intensive care, with progressively increasing requirements of inotropes, mechanical ventilation and broad-spectrum antibiotics, the patient succumbed to multi-organ dysfunction syndrome on post-operative day 6.

Histopathological report was as follows:

Ovary-oedematous ovarian stroma with few thick-walled blood vessels, few follicles, tiny foci of fresh haemorrhages. Biopsies of omentum and peritoneum showed no evidence of atypia or malignancy

Discussion

Massive ovarian oedema as a clinical entity is still poorly understood, with myriad presentations, lack of pathognomonic features on imaging modalities and no clear consensus on management till date. Primary oedema occurs in an otherwise healthy ovary and is generally thought to be the outcome of repeated torsion hampering venous and lymphatic drainage, without affecting arterial supply, resulting in stromal oedema often with cystic changes [6]. Secondary oedema occurs as a consequence of an ovarian or extra-ovarian pathology which may be neoplastic (ovarian tumours, metastasis), non-neoplastic (PCOS, fibromatosis) or even iatrogenic (ovulation induction) [6].

As per a review of the existing literature on this entity in 2013 by Parveen *et al.*, 85% of cases of massive ovarian oedema were primary and 15% secondary. A large majority of cases (89%) were managed by either salpingo-oophorectomy or abdominal hysterectomy with bilateral salpingo-oophorectomy. Only 11% cases underwent fertility-sparing procedures. In this small subset (20 out of 177 cases) only 5 patients were subjected to totally non-invasive management (radiological monitoring with symptomatic treatment). Overall, out of 177, only 76 cases (43%) showed intraoperative evidence of ovarian torsion [7].

Many case reports suggested that a large number of such cases would respond to judicious wedge resection of the affected ovary and symptomatic management and decision for extirpative surgery should be made after carefully weighing risks v/s benefits and thorough patient counselling [8-10].

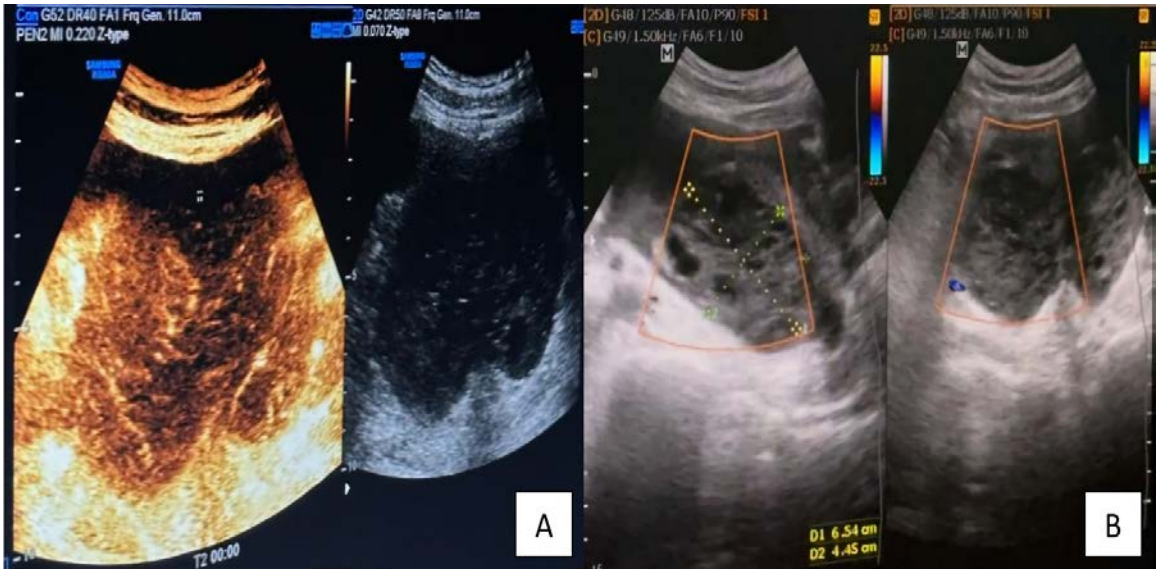

Fig 1: (A) Contrast enhanced ultrasound showing delayed time to peak, (B) Ultrasound of ovary showing stromal oedema

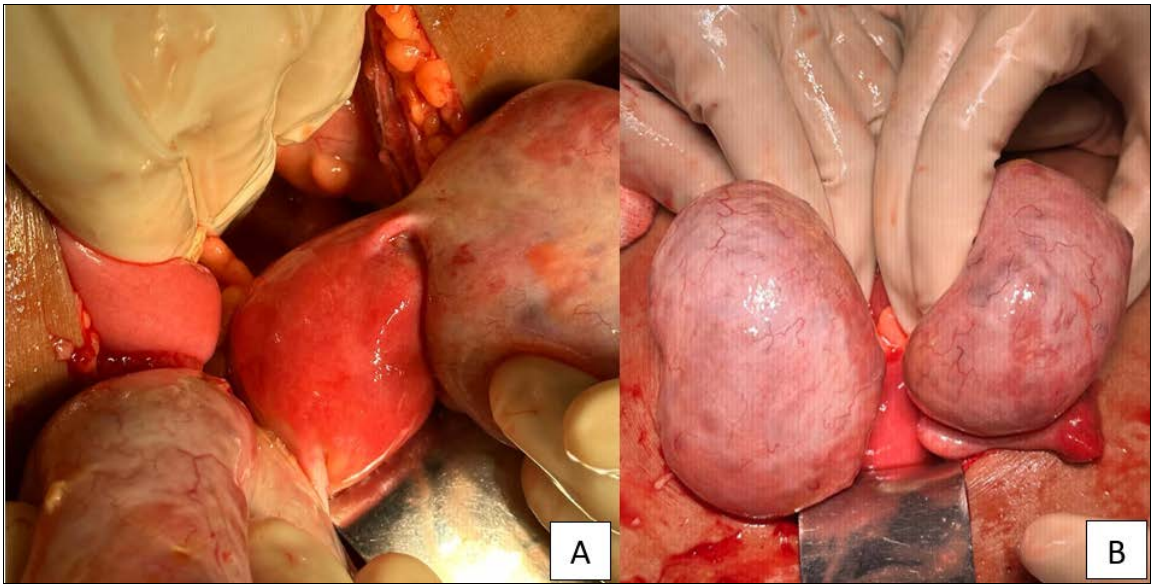

Fig 2: (A) Intra-operative findings showing normal uterus with bilateral bulky ovaries, (B) Bilateral bulky ovaries with no evidence of necrosis or torsion

In our case, the patient was a young girl (19-year-old) who presented with features of acute abdomen, radiological imaging strongly suggestive of bilateral ovarian torsion and clinical deterioration with symptomatic treatment. Hence, a decision was made for exploratory laparotomy, which showed no evidence of torsion or ovarian necrosis, and all the biopsies (ovarian, omental, peritoneal) were negative for evidence of malignancy. Hereby, our case report reiterates the chameleon-like nature of this mysterious clinical entity and calls for further research into the aetio-pathology and appropriate management algorithms.

Conclusion

Massive ovarian oedema is a rare clinical entity that generally masquerades as an ovarian mass in reproductive age, often as acute abdomen mimicking ovarian torsion or appendicitis or more indolent with radiological findings casting the ominous suspicion of a malignancy. Owing to a large majority of cases being benign, and the prevalence of over-treatment, it is essential that clinicians remain wary and ensure optimum treatment whilst balancing the preservation of hormonal function and fertility.

Conflict of interest: The authors declare no conflict of interest.

Funding: None.

Compliance with ethical standards: The case report was published with valid informed consent from the patient's parents. Consent was also taken to publish intra-operative images for academic purposes.

Ethical clearance: Not required.

References

1. Roth LM. Massive ovarian edema with stromal luteinization: A newly recognized virilizing syndrome apparently related to partial torsion of the mesovarium. Am J Clin Pathol. 1971;55:757-60.
2. Kalstone CE, Jaffe RB, Abell MR. Massive edema of the ovary simulating fibroma. Obstet Gynecol. 1969;34(4):564-71.
3. Geist RR, Rabinowitz R, Zuckerman B, Shen O, Reinus C, Beller U, *et al*. Massive edema of the ovary: A case report and review of the pertinent literature. J Pediatr Adolesc Gynecol. 2005;18(4):281-4.
DOI: 10.1016/j.jpag.2005.05.008.
4. Varma A, Chakrabarty P, Gupta G. Massive ovarian edema: A case report presenting as diagnostic dilemma. J Family Med Prim Care. 2016;5(1):172-4.
5. Roberts CL, Weston MJ. Bilateral massive ovarian edema: A case report. Ultrasound Obstet Gynecol. 1998;11(1):65-7.
6. Lakshmi KA, Johnson T. Massive edema and fibromatosis of ovary: A rare case report. Indian J Pathol Oncol. 2024;11(1):87-9.
7. Praveen RS, Pallavi VR, Rajashekar K, Usha A, Umadevi K, Bafna UD, *et al*. A clinical update on massive ovarian oedema: A pseudotumour? Ecancermedicalscience. 2013;7:318.
8. Akshaya PKKA, Charumathi R, Madhumitha J, Vijayaraghavan B. A rare case report of massive ovarian edema. Int J Reprod Contracept Obstet Gynecol. 2022;11:2042-5.
9. Young RH, Scully RE. Fibromatosis and massive edema of the ovary, possibly related entities: A report of 14 cases of fibromatosis and 11 cases of massive edema. Int J Gynecol Pathol. 1984;3(2):153-78.
10. Daboubi MK, Khreisat B. Massive ovarian oedema: Literature review and case presentation. East Mediterr Health J. 2008;14:972-7.

How to Cite This Article
Rambhia KK, Banerjee Y. A rare case of bilateral massive ovarian oedema masquerading as torsion. International Journal of Clinical Obstetrics and Gynaecology 2024;8(4):131-133.