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CASE REPORT

Rare Case of Bilateral Ovarian Lymphangioma with Chylous Ascites in Pregnancy with Review of Literature

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Case

A 36-year-old primigravida underwent elective caesarean section at 38 weeks of gestation for breech presentation. She had conceived after in vitro fertilization (IVF) done for poor ovarian reserve (Sr. Anti-mullerain hormone < 0.03 and Antral follicular count-3) with donor oocytes. Her antenatal course was uneventful. During her caesarean delivery, upon opening the peritoneum, chylous ascitic fluid was noted in the abdomen. The fluid was collected and sent for routine microscopy, cytology and culture sensitivity testing. Around 400 ml of chylous ascitic fluid was drained. She delivered a healthy baby boy (3 kg) with good APGAR score. The uterus and both fallopian tubes appeared normal. However, bilateral ovaries were bulky (right ovary-8 × 7 × 5 cm, left ovary-7 × 6 × 5 cm) with chylous fluid oozing from both the ovaries (Fig. 1). Husband and relatives were explained about the intraoperative findings. After taking their consent, bilateral ovarian tissue biopsies was taken and sent for histopathology. Other organs like the stomach, pancreas, jejunum, mesentery, etc. did not show any abnormality or growth. An abdominal drain was kept. The drain had 200 ml of milky serosanguinous fluid on the first post-operative day, which reduced to less than 20 ml on fifth day. The patient was discharged after removal of abdominal drain and advised to follow-up after 7 days. Her investigations showed the following results as seen in Table 1.

However, on day 9 of delivery, she came with breathlessness and severe abdominal distension. Her vitals were stable with oxygen saturation of 96% and reduced air entry on the right lower side of chest with ascites. Her chest x-ray revealed moderate pleural effusion on the right side. Ultrasonography of the abdomen and pelvis showed moderate ascites with enlarged ovaries suggestive of lymphangiomas. The patient was readmitted. Around 3 l of chylous ascetic fluid was removed and another 500 ml on pleural tapping. These samples were sent for further investigations which confirmed chylous fluid (Table 2). The patient was given intravenous albumin and antibiotics. She was symptomatically better and discharged 2 days later.

The histopathology of ovarian biopsies revealed multiple dilated channels in ovarian parenchyma with some containing eosinophilic fibrinous fluid material (Fig. 2). The channels were lined by uniform flattened endothelial cells and separated by moderate cellular eosinophilic stroma suggestive of bilateral ovarian lymphangiomas with differential diagnosis of hemangiomas or adenomatoid tumours (Fig. 2). The immunohistochemistry showed ovarian tissue lining endothelial cells positive for CD31 and focal weak positivity for D2 40, while equivocal for CD34. The cells were cytokeratin and vimantin negative, with no mitotic activity, which was consistent with diagnosis of bilateral ovarian lymphangiomas (Fig. 2).

The patient had recurrence of symptoms after 3 weeks and presented with breathlessness and abdominal distension. On examination, her vitals were stable, with oxygen

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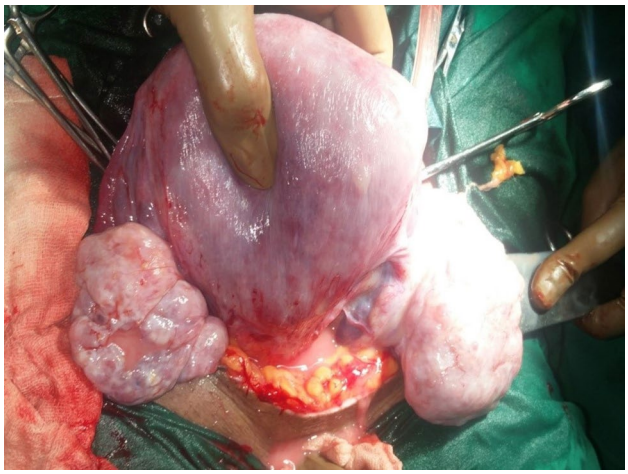

Fig. 1 Chylous fluid oozing from both ovaries

saturation of 94%, severe ascites and reduced air entry on the right side of chest. X-ray chest showed right-side moderate pleural effusion, and there was moderate to severe ascites on ultrasonography. In view of recurrence of symptoms, a decision for exploratory laparotomy was taken with the consent of the patient and her husband. Intra-operatively, 3 l of chylous ascetic fluid was drained. Both ovaries were enlarged and oozing chylous fluid. Bilateral oophorectomy was done and an abdominal drain kept. The other common sites for lymphangiomas like stomach, jejunum, mesentery etc. were thoroughly checked for any growth or abnormalities. Post-operative course was uneventful and she was discharged on day 5. The patient was asymptomatic on 6-month follow-up.

Discussion

Extravasation of milky chyle rich in triglycerides into peritoneal cavity leads to chylous ascites. True chylous ascites is characterized by high fat (triglyceride > 110 mg/dl). Secondary chylous ascites is seen when existing clear ascitic fluid turn chylous. The literature review shows that chylous ascites during pregnancy is an extremely rare occurrence and could be due to various causes like pancreatitis, inflammation, lymphangioma, etc. (Table 3) [1, 2].

Lymphangiomas are rare benign congenital malformations of lymphatic system, but less than 1% may turn malignant [1]. Its pathogenesis is not very clear and is rarely seen in adults. Lymphangiomas occur due to obstruction of local lymph flow, and in 95% of people, it's found in the head, neck and axillary region [1, 2]. Only 5% may have involvement of lungs, pleura, pericardium, oesophagus, stomach, jejunum, colon, pancreas, liver, gallbladder, kidney and mesentery [1, 2]. Primary ovarian lymphangioma is extremely rare with only around 20 cases

Table 1 Investigations done on the chylous ascitic fluid

Sr. no.	Investigation	Result
1	Chylous ascitic fluid for cytology	Polymorphonuclear leucocytes and few lymphocytes. No malignant cells
2	Chylous ascitic fluid for routine microscopy	Specific gravity is 1.03 Total fat content is 20 g/l Glucose and amylase levels-normal Triglycerides > 120 mg/dl Cholesterol level-low Total protein-3.9 g/dl
3	Chylous ascitic fluid for culture report	No growth No AFB seen

Table 2 Investigations done on the chylous ascitic fluid and pleural effusion

Sr. no.	Investigation	Result
1	Chylous fluid for cytology	Polymorphonuclear leucocytes and few lymphocytes. No malignant cells
2	Chylous fluid for routine microscopy	Chylous fluid Specific gravity is 1.05 Total fat content is 25 g/l Glucose and amylase levels-Normal Triglycerides > 130 mg/dl Cholesterol level-low Total protein-3.4 g/dl
3	Chylous ascitic fluid for culture report	No growth No AFB seen

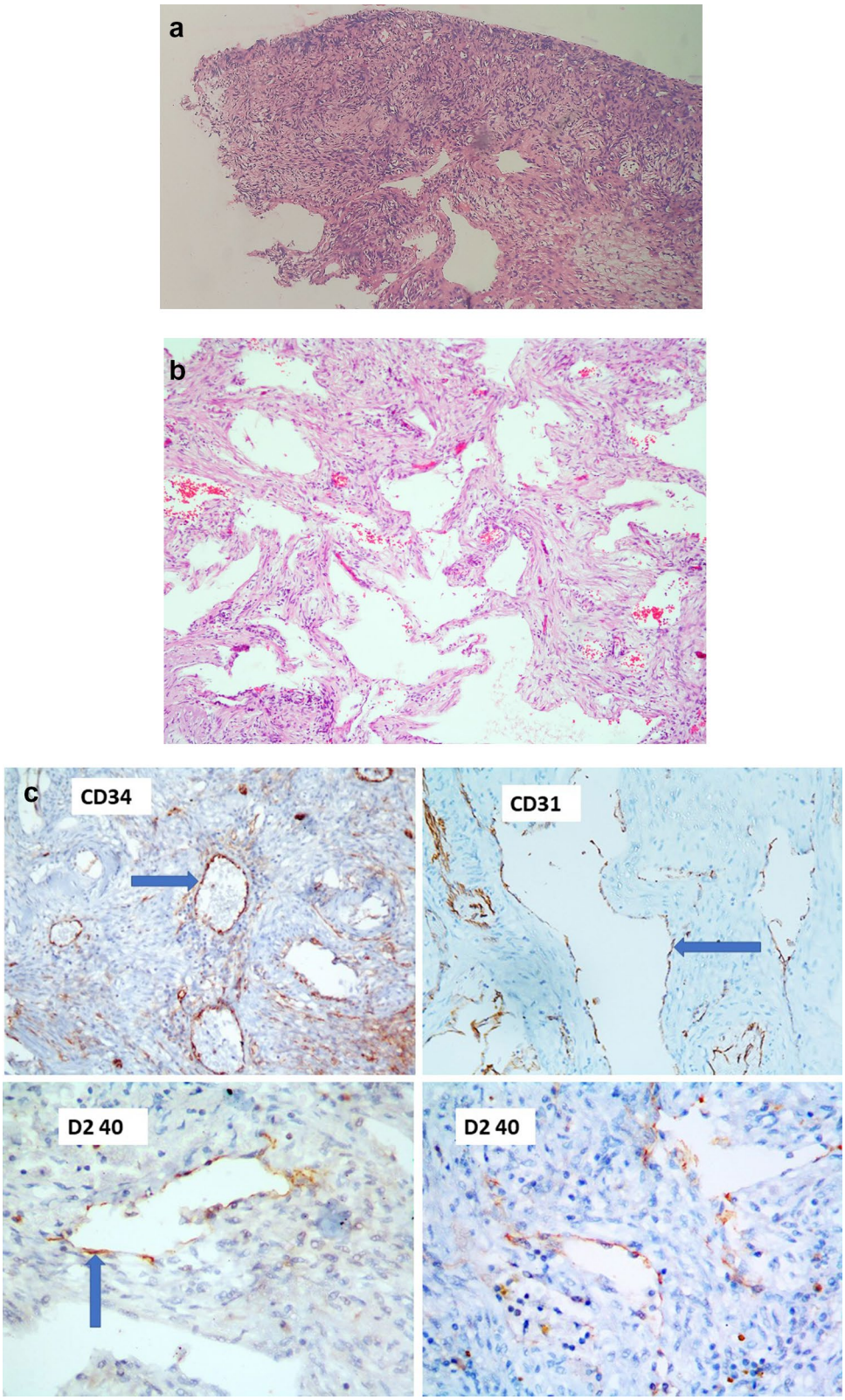

Fig. 2 Histopathology and Immmunohistochemistry of the ovaries

reported in the literature [1–4]. Three of them had milky ascites with ovarian lymphangioma [1–4]. One woman presented with 33 weeks of gestation with ovarian lymphangioma and torsion [1–4]. Most women were managed with total abdominal hysterectomy with bilateral salphino-ooprectomy (TAH BSO) [1–4]. There were only 3 case reports (non-pregnant women)

Table 3 Cases presenting with chylous ascites and pregnancy

Sr no.	Year	Author	Presentation	Final diagnosis
1	1980	Kondrat'ev et al.	Volvulus of small intestine	Volvulus
2	2001	Liu et al.	Pancreatitis Resolved post-operatively	Pancreatitis
3	2005	Habek et al.	Chylothorax secondary to pulmonary TB in childhood	Congenital lymphangiectasia
4	2006	Chuang et al.	Acute abdomen, severe pancreatitis	Pancreatitis
5	2007	Sun et al.	Incidentally at LSCS Mesenteric tumour (spiral CT abdomen and pelvis)	Mesenteric fibromatosis
6	2012	Babic et al.	Spontaneous chylous ascites, morbid obesity	Complete resolution, no malignancies noted on CT
7	2016	Daddenavar et al.	Pregnancy induced hypertension with ascites and respiratory distress	Bilateral ovarian cystic lymphangioma

of ovarian lymphangiomas with chylous ascites who underwent unilateral or bilateral oophrectomy [1, 2, 4]. There is only one reported case of pregnant woman with ovarian lymphangioma and chylous ascites like in our patient. She presented at 37 weeks of gestation with breathlessness and pre-eclampsia and underwent caesarean delivery with unilateral ovarian oophrectomy for an enlarged left ovary (16 cm) [3].

Conclusion

Our case highlights the importance of diagnosis of primary ovarian lymphangioma in women presenting with unilateral or bilateral ovarian enlargement with chylous ascites. The diagnosis is uncertain preoperatively as little known about its clinical behaviour. In our patient, we had to do ascetic and pleural tapping as she presented with severe respiratory distress. The diagnosis was confirmed with immunohistochemistry. When reviewed retrospectively, removal of the ovaries during caesarean section could be recommended in such cases to prevent morbidity.

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Compliance with Ethical Standards

Conflict of interest There is no conflict of interest for the manuscript submitted.

Human and Animal Rights The manuscript submitted for consideration does not involve any research or experiments on human participants and/or animals.

Informed Consent Informed consent was taken from the patient and her husband before submitting the manuscript for consideration to this journal.

References

1. Iwasa T. Lymphangioma of the ovary accompanied by chylous ascites. J Obstet Gynaecol Res. 2009;35(4):812–5.
2. Al-Busafi SA, Ghali P, Deschênes M, et al. Chylous ascites: evaluation and management. ISRN Hepatol. 2014;2014:240473. https://doi.org/10.1155/2014/240473.
3. Daddenavar A, Daddenavar V, Patil R, et al. Bilateral ovarian cystic lymphangioma with chylous ascitis in pregnancy—a rare case report with review. Int J Reprod Contracept Obstet Gynecol. 2016;5(5):1633–6.
4. Choudhary RA, Vora PH, Deodhar KK, et al. Bilateral lymphangioma of ovary—a rare case report. Sch J Med Case Rep. 2016;4(10):723–5.

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