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A Rare Case of Recurrent Mucinous Cystadenoma in Pregnancy

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Abstract: ***Ovarian mucinous cystadenomas are second most common type of benign epithelial ovarian tumors. They may be intra peritoneal or retroperitoneal, of which intraperitoneal tumors are commonly seen while primary retroperitoneal mucinous cystadenomas (PRMCs) are very rare. Our is a case of a 26-year-old pregnant woman in whom recurrent ovarian cystadenoma was operated successfully. Mucinous cystadenoma recurrence is apparently not as rare as reported in the literature. The unique feature in our case, was that patient had fourth time recurrence of mucinous cystadenoma, despite undergoing, twice laparoscopic and once laparotomy removal of cyst on previous three occasions. The recurrence may have been due to intraoperative cyst rupture or conduct of cystectomy during previous surgeries. Also in our case, the recurrent tumor was retroperitoneal in nature which may have been because of two possibilities: first, the tumor may have been intraperitoneal at first onset and became retroperitoneal during recurrence due to cyst rupture and spillage of mucinous material in the cavity during previous surgeries; second, the tumor may have been primarily retroperitoneal, although a rare phenomenon. The incidence for primary retroperitoneal cystadenoma is around 10 for every one million hospital admissions.***

Keywords: Mucinous Cyst Adenoma, Pregnancy, Pre-Eclamsia, Retro-Peritoneal

1. Introduction

Ovarian mucinous cystadenoma is a benign tumour arising from the ovarian surface epithelium and is the second most common type of epithelial ovarian tumors. [1, 2]

Of all ovarian tumours, mucinous tumours comprise 15%; of these mucinous tumours around 80% are benign, 10% are borderline while 10% are malignant. [3, 4] It is usually unilateral, only 10% are bilateral. [5] Mucinous tumours are characterized by multilocularity, smooth outer and inner surfaces. [2] Complications that are commonly seen with benign ovarian cysts in general are torsion, haemorrhage and rupture. [6] Since these tumors are filled with mucinous fluid, rupture leads to mucinous deposits on the peritoneum. These tumors are rarely seen in adolescents or in pregnant females. [5, 7] During pregnancy the frequency of ovarian tumours is around 1 in 1,000 pregnancies, malignant tumors are seen in 1 out 15,000- 32,000 pregnancies. [2] During pregnancy of all the adnexal masses 28% are serous or mucinous cystadenomas.

Our patient showed a recurrence with Mucinous cystadenoma. The biologic behavior of mucinous ovarian tumors depends on the specific histologic variant and stage. Intraepithelial (non-invasive) mOC, FIGO stage I, has a recurrence rate of only 5.8 %.

Ovarian cystadenomas may be intra peritoneal or retroperitoneal. Intraperitoneal tumors are commonly seen. Primary retroperitoneal mucinous cystadenomas (PRMCs) are very rare as indicated by the small number of cases reported in the literature. Most patients are diagnosed without specific symptoms, but, like most retroperitoneal obstructive effect on adjacent organs. Once diagnosed, the tumor should be completely removed because of the risk of infection, recurrence and malignant degeneration. Approximately 50 cases of primary retroperitoneal mucinous cystadenocarcinomas have been reported in the international literature, but only 29 cases of a PRMC have been described [10].

2. Case Report

A 26-year-old G2P1L0 with a history of eight months of amenorrhea was referred to Dhiraj general Hospital on 24th Nov 17 for pregnancy induced hypertension, blurring of vision and with ultrasonography reports suggestive of right sided ovarian cyst. On medical history evaluation, patient complained of blurred vision in the last 24 hours which had gradual onset and was intermittent in nature. Patient had no complain of either pain in abdomen, bleeding or leaking per vaginum. Menstrual history revealed that she had regular menstrual cycle of 28 to 30 days and menses lasted for 3-4 days. Date of last menstrual period of 16 April 2017 and therefore the expected date of delivery was 23 Jan 2018 and gestational age at the time of presentation of complaints was 31 weeks and 6 days.

Obstetrics history revealed that she was gravida – 2, para – 1 and live birth – 0. The first pregnancy had occurred 4 years back, it was preterm vaginal delivery after 8 months of amenorrhea and was associated with intrauterine death. Current pregnancy was 2nd gravida. Two tetanus toxoid injections were received by patient and was taking iron, folic acid and calcium supplements.

Patient had undergone three surgeries for serous mucinous cystadenoma. She underwent laproscopic surgery for serous cystadenoma of right side in 2013, laproscopic surgery for mucinous cystadenoma of right side in 2014 and laparotomy for mucinous cystadenoma of right side in 2015.

She was a non-addict having normal sleep and appetite, bowel and bladder habits were also normal.

Patient was oriented to time, place and person. Pallor was absent; however, pedal edema was present. Vitals assessment revealed that patient had hypertension with a blood pressure of 140/100 mm of Hg, pulse was 90 beats per minute and oral body temperature was normal. Urine dip stick revealed proteinuria of +2. Other systems were found to be normal.

Per abdomen examination revealed that abdomen was relaxed. Size of uterus correlated with 32 weeks of pregnancy. Presentation was oblique, with fetal head on maternal left side. Fetal heart sounds were present on auscultation on maternal left side, they were regular. Patient was admitted for further investigations and tablet labetalol in the dose of 100mg twice daily was started to control blood pressure.

Blood investigations revealed following: Blood group was AB positive, sickling negative, hemoglobin 9.2gm/dl, total white blood cell count was 19,000/ul, total platelets were 3.47lac/ul, random blood sugar was 104mg/dl, serum urea was 84mg/dl, serum creatinine was 2.0mg/dl, SGOT was 44IU/L, SGPT was 36 IU/l, ALP was 42u/l, CA-125 – 138.4 U/l, T3- 1.12ng/l, T4 – 12.0µg/dl and TSH – 7.97µIU/ml, urea/creatinine spot/24hour was 78mg/dl, Serum protein was 6gm/dl, with albumin and globin being 3gm/dl each, APTT was 30 sec, PT was 14 sec and INR was 1.0, uric acid was 7.89mg/dl, LDH was 1254U/l, Serum total bilirubin was 1.0mg/dl (direct – 0.4mg/dl and indirect – 0.6mg/dl), serum electrolytes were sodium 134mmol/l, potassium 4.6mmol/l and chloride 105mmol/l.

Patient was referred to medicine department for further evaluation, where the patient was advised to start inj ceftriaxone 1gm twice daily, inj metronidazole thrice daily and tablet nifidipine 20mg once daily and blood pressure monitoring 2 hourly. Ophthalmology reference revealed no significant abnormality.

Antenatal ultrasonography on 17 July 2017 revealed single live intrauterine fetus of mean maturity of 14 weeks by BPD, 13 weeks 3 days by CRL, and13 weeks 4 days by FL and 13 weeks 1 day by AFA. Expected date of delivery was 18th Jan 2018. Fetal movement and heart sound were present. Placenta was fundo-anterior. Uterine Doppler flow was normal. No gross abnormality was present. Loculated right ovarian cyst 16.6cm*8.9cm in size with hydronephrotic right kidney. Uterus was displaced to left side.

Antenatal ultrasonography on 24 Nov 2017 revealed single live intrauterine fetus of mean maturity of 29 weeks by BPD, 26 weeks and 6 days by AC, 27 weeks and 3 days by FL and 27 weeks and 5 days by AFA. Estimated fetal weight was 1069gms and expected date of delivery was 18th Feb 2018. Fetal movement and heart sound were present. Placenta was anterior, more towards maternal left. Severe oligohydromnios was present with amniotic fluid index was 4.5 and MVP was 2.8. Presentation was oblique. No gross congenital malformation was present. complex multicystic lesion in right adnexa with thick internal septations within measuring 12.7x 10.7 cm with internal vascularity. Liver, gall bladder, portal vein, common bile duct, spleen and pancreas were normal, right kidney showed gross hyronephrosis with dilated proximal ureter. Moderate intraperitoneal free fluid was also seen.

MRI pelvis revealed large, multi-septated cystic lesion in right adnexal region, extending in lower abdomen, upto umbilical level with mass affect over uterus and urinary bladder and compression over right ureter with mild to moderate proximal hydro nephrosis and hydroureter. Most likely benign right ovarian cystic neoplasm.

Patient was kept on expectant management with close observation on mother and fetus till she reaches term. But on 26th Nov 17 fetus died in utero.

On 27th Nov 17, Laprotomy followed by preterm lower segment caesarean section (lscs) followed by right sided cystectomy in case of right sided ovarian mass. Intra-peritoneal adhesions were removed by adhesiolysis. Ovarian cyst was sent for cytology. Cytology reports revealed mucinous papillary cystadenoma.

3. Discussion

The average incidence of adnexal masses with pregnancy, is 1 in 200 pregnancies. Only a few cases of ovarian mucinous cystadenoma with pregnancy have been reported in the literature. Early diagnosis of mucinous cystadenoma is important because of its malignant potential.

This is the rare case of recurrent, large mucinous benign cyst seen in a pregnant woman, which had spread & extended retro-peritoneally. In our case, a multi loculated right ovarian mucinous cystadenoma measuring 12.7x 10.7 cm at 31 weeks of pregnancy was seen with pre-eclampsia and IUGR.

Various studies have reported variety of presentation, in sizes.The size of the cyst seen in a study by Mummigatti KA et al., was 20.5x13.8x16cm. [2] Qublan et al., described a 6300g multi loculated right ovarian mucinous cystadenoma showing stromal luteinization and measuring 33x24x20cm at 38 weeks of pregnancy. [11] Literature suggest that if adnexal masses are present during pregnancy, surgical intervention should be avoided due to adverse fetal outcomes as a result of preterm delivery or premature rupture of membranes. [12]

In our case, an emergency surgical intervention was not needed as there was no immediately pressing reason, or unbearable abdominal symptoms. However, as the fetus died in utero, surgical intervention was carried out.

The unique feature in our case, was that patient had fourth time recurrence of mucinous cystadenoma, despite undergoing, twice laparoscopic and once laparotomy removal of cyst on previous three occasions. This may be due to only cystectomy done in last three surgeries rather than oophorectomy, as in our case patient was young, and it was desirable to preserve her fertility.

In the study by Ben-Ami I et al, the authors tried to evaluate the cause of recurrent benign ovarian mucinous cystadenoma. They observed that mucinous cystadenoma recurrence is apparently not as rare as reported in the literature. They proposed a theory, that intraoperative cyst rupture and cystectomy are the two risk factors responsible for recurrence. [13]

In our case, there was recurrence, with intra-peritoneal adhesions. Adhesiolysis was done. It was possibly due to cyst rupture and spillage of mucinous material in the cavity, during previous surgeries, which may well be a reason for Retro-peritoneal extension of the mucinous cyst adenoma, a feature hitherto not found on laparotomy, of many cases.

Another possibility for retroperitoneal extension is that the tumor was primarily retroperitoneal in nature. Retroperitoneal cysts are rare and the accurate incidence is unavailable. The histogenesis remains unclear, although four main hypotheses have been advanced with regard to the formation of retroperitoneal mucinous tumors. According to the first three hypotheses, the tumor arises either from ectopic ovarian tissue, from a teratoma in which the mucinous epithelium has overridden all other components to survive as a single cell component, or from remnants of the embryonic urogenital apparatus. Recently, a fourth theory has gained wide acceptance. This theory suggests coelomic metaplasia as the causal agent, whereby tumors arise from invagination of the peritoneal mesothelial layer that undergoes mucinous metaplasia with cyst formation.

There were some other cases of mucinous cystadenoma in past which were operated in our hospital at 14-16 weeks of gestational age, large enough to occupy most of the space in abdominal cavity & cramp the foetal growth. The cyst was removed to allow unhindered growth of foetus, and after that their pregnancy was uneventful.

4. Conclusion

Mucinous cystadenoma recurrence is apparently not as rare. When the surgeon enters the abdomen, care should be taken to remove the involved ovary intact without spillage of the mucinous contents, as rupture may increase its potential for recurrence, oophorectomy instead of cystectomy should be preferred to decrease chances of recurrence. In a pregnant female, surgery should be delayed till full term is reached, as usually the pregnancy is not disturbed in most of cases. Surgery to be performed during puerperial period, as surgical emergency like torsion is common after delivery.

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