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Case Report

Luteoma in pregnancy: a rare cause of threatened preterm labour!

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ABSTRACT

Pregnancy luteoma is a rare non-neoplastic tumor-like mass of the ovary. They are usually asymptomatic and found incidentally during ultrasound imaging or surgery. Rarely do they present with pain abdomen mimicking threatened preterm labor. They regress spontaneously after delivery. We presented a case of a 29-year-old G4A3 with twin pregnancies and chronic hypertension who presented with acute flank pain, constipation, and occasional hardening of the uterus. A provisional diagnosis threatened preterm labor with suspected ureteric colic and cystitis was made. The patient was initially managed on the same line but on a detailed in-patient evaluation her ultrasound revealed bilateral multicystic ovaries with few hypoechoic areas inside it mostly suggestive of 'luteoma of pregnancy'. The patient had acne, hirsutism, and chronic hypertension well controlled on antihypertensives. The patient delivered twins successfully by cesarean section and luteoma and symptoms were resolved postpartum after 4 months. Recognition of this entity is important so that malignancy can be ruled out and unnecessary surgery, with concomitant risk to both the mother and the fetus, is avoided.

Keywords: Luteoma, Pregnancy, Non-neoplastic tumor

INTRODUCTION

Pregnancy luteoma is first described by Sternberg and Barclay in 1966. In 1963, Sternberg described a solid ovarian tumor that developed during pregnancy and was composed of large acidophilic luteinized cells. These presented as an exaggerated luteinization reaction of the normal ovary.[1] The size of these cysts were most frequently between 5 and 10 cm, however, there are some reported pregnancy luteoma cases which are extremely large (up to 20 cm).[2] Some women can suffer from general symptoms like pelvic pain, lumbalgia and constipation. Associated with acne, hair loss, hirsutism, clitoromegaly, deepening of voice and virilization symptoms.[3] Usually, pregnancy luteoma is asymptomatic condition and diagnosed incidentally during antenatal scans or intraoperatively at time of caesarean section or post-partum tubal ligation.[4]

CASE REPORT

A 29-year-old patient with gravid 4 and previous 3 abortions at 29 weeks of pregnancy with spontaneous conception, recurrent pregnancy loss and chronic hypertensive dichorionic diamniotic twin gestation presented to obstetrics and gynecology department of Institute of Kidney Diseases and Research Centre, Ahmedabad with pain abdomen-right flank radiating to lower abdomen, constipation and occasional tightening of uterus associated with 2 episodes of vomiting. Patient had fever one day after admission. There was no history of dysuria, bleeding or leaking per vaginal. She perceived adequate fetal movements. Obstetric history was 1st abortion occurred at 3 months, second at 2 and half months and third at 5 months because of cervical incompetence. Surgical history was at 16 weeks, in this pregnancy, history indicated cervical encirclage was done. Medical history,

she had history of chronic hypertension in the past 2 years. She was on medications labetalol 100 mg thrice a day and nifedepine 20 mg thrice a day. Family history was nothing significant.

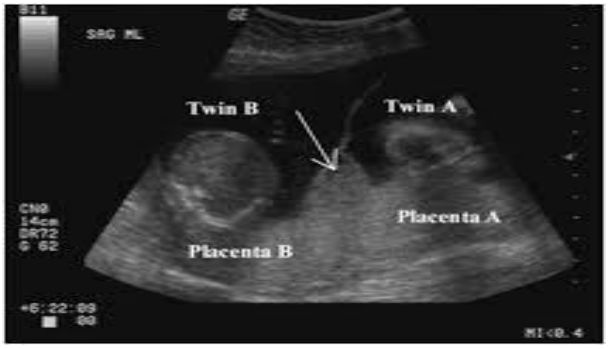

Figure 1: Dichorionic diamniotic twins.

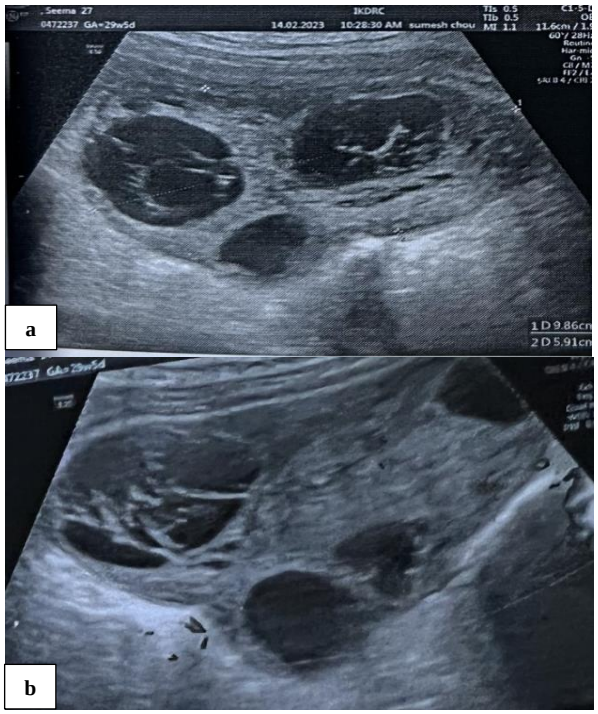

Figure 2 (a and b): Ultrasonography image of bilateral luteoma in pregnancy.

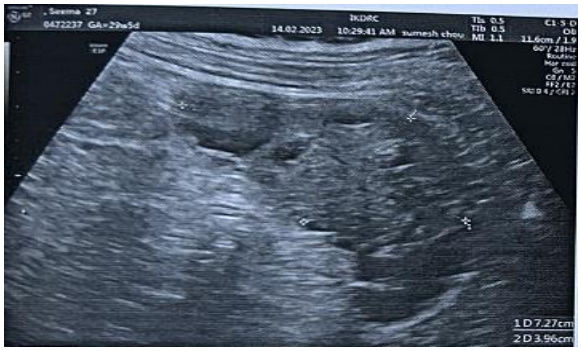

Figure 3: Solid component of 7.27×3.96 cm.

Figure 4: Luteoma in pregnancy during LSCS.

On general examination patient was obese with acne, hirsutism. There was no pallor, icterus, cyanosis, clubbing, lymphadenopathy. Vitals showed afebrile, pulse=100 beats/min, blood pressure=130/80 mmHg. On obstetric examination: uterus 33 weeks, 2 contraction in 10 min for 20-25 sec duration, multiple fetal parts palpated, two distinct fetal heart sounds auscultated and confirmed with hand-held Doppler. Right iliac fossa tenderness present.

Routine blood investigations including complete blood count, liver function tests, renal function tests, oral glucose tolerance test were within normal limits. 24 hour urine protein volume was 2950 ml, 24 hours urine protein was 0.16 gm/24 hours, spot urinary creat was 85.78 mg/dl and spot urinary protein was 17.2 mg/dl. Protein:creatinine ratio was 0.20. Urine routine showed 40-50 /high power field pus cells and urine culture report showed growth of *Enterococcus fecalis* >1 million cfu/ml resistant to antibiotics. Sr. ANA was 0.142 index (range=0-1), S. DSDNA was 10.8 IU/ml range (0-25 IU/ml), P-ANCA 1.05 U/ml and C-ANCA 1.04 U/ml (rang=0.01-5 U/ml), uric acid was 3.8 mg/dl, S.C3 was 110 (range 90-207 mg/dl), S. C4 was 37.3 (range=17.4-52.2 mg/dl). Additional investigations were done. Androgen levels were borderline high, serum testosterone level=1.8 ng/ml (normal range: 0.5-0.8 ng/ml). Alpha feto protein=200 ng/ml (160-550), LDH=150 U/lit (140-280), CA 125=25 U/ml (<35), CEA=2.6 mcg/lit (2.5-5), CA19=9-10 U/ml (<40), inhibin=40 pg/ml (<45). USG KUB was done which was normal.

Ultrasonography was suggestive of twins, live, intra-uterine, Dichorionic diamniotic gestation of date 30.6 weeks, corresponding to 29.3 weeks and 28.6 weeks respectively with normal growth and morphology.

Bilateral ovarian size was increased. Size of right ovary was 9.86×5.91 cm and size of left ovary was 9.54×6.1 cm. Multiple cysts seen in bilateral ovary with few hypoechoic areas inside it mostly suggestive of 'luteoma of pregnancy'. No evidence of torsion was noted.

Injection ceftriaxone was given 1 gm intravenously 12 hourly. For pain injection paracetamol was given. Her repeat 24 hour urine protein was done which was 0.05 g/24 hours. Fosfomycin sachet 3 g with 200 ml of water was given for 4 weeks. She had negative urine culture report

done after a week. After one week of antibiotics patient was not relieved of symptoms. Patient was treated conservatively and decision was taken to monitor patient with subsequent periodic monthly follow-ups with ultrasonography and androgen levels if required till 3 months after delivery. There was no obvious increase in size of luteoma of pregnancy was noted. Also, there was no obvious increase in androgen levels.

Table 1: Differential diagnosis of ovarian tumors.

Type of tumor	Age (years)	Lab findings	Imaging features	MRI	Histopathology	Gold standard
Theca lutein cysts	15-45	Depends on the underlying e tiology	In US we may see bilaterally enlarged ovaries with multiple cysts	Multiple bilateral cysts	Theca interna cell Hyperplasia	History/ imaging
Sertoli leydig cell tumors	15-35	Elevated ser um testosterone level Elevated alp ha-fetoprotein	In US we may see unilateral Well-defined hypoechoic lesion	Low T2 signal intensity areas of high signal intensity	Leydig cells (Polygonal pink cells with eosinophilic cytopl asm) Sertolicells (clear vacuolated cytoplasm)	Biopsy
Granulosa cell tumours	50-60	High level of estrogen and progeste ron We may see inhibin, calretinin, and Ki-67 on the surface of granulosa tumor cells	In US - Large multilocular solid and cystic mass with areas of hemorrhage, sponge like app,associated with endometrial thickening or endometroid carcinoma	We may see solid, cystic, or multiloculated solid and cystic mass	Call Exner bodies	Biopsy
Dysgerm inomas	20-30	High level of HCG and LDH Hypercalce mia	Large solid unilateral mass with fibrovascular septa and relatively homogenous areas, speckled calcifications	We may see ovarian mass with septation which are hyperintense on T1 and hypo or isointense on T2 imaging	Sheets fried egg appearance cells	Biopsy
Yolk sac tumours	Young children, male infants	High levels of AFP	Large unilateral solid and cystic mass with hemorrhagic areas. Dilated blood vessels(bright dot sign)	Ovarian mass with hemorr hagic areas	Yellow appearance Hemorrhagic Schiller-Duval bodies (glomeruli like structures)	Biopsy
Serous cystaden oma	>55	Elevated levels of serum cancer antigen-125	In US we may see simple or multiloculated cy st In serous cystadenocarcino ma we may see papillary proj ection inside the cyst	We may see a simple cyst with beak sign, hypointense on T1 and hyperintense on T2 We may see some Solid malignant c omponents inside the cyst with	Cyst wall consist of benign/malignant Fal lopian epithelial layer Psammoma body We may see papillary projection inside the cyst	Biopsy

Continued.

Type of tumor	Age (years)	Lab findings	Imaging features	MRI	Histopathology	Gold standard
			In serous cystadenocarcinoma we may see ascites	intermediate signal on T1 and T2		
Thecoma	>50	High level of estrogen	In US we may see non-specific ovarian mass We may see evidence of endometrial hyperplasia due to increased level of estrogen	Hyperintense on T2 T1 intensity depends on the amount of fibrous tissue (fibrous tissue lead to hypointensity)	Lipid-laden stromal cellswith pale, vaculolated cytoplasm	Biopsy
Teratoma	10-30	High level of HCG and LDH	In US we may see cystic adnexal mass with mural components and echogenic lesion due to calcification The iceberg sign Dot-dash pattern	We may see evidence of fat components	All three germ layers cell	Biopsy

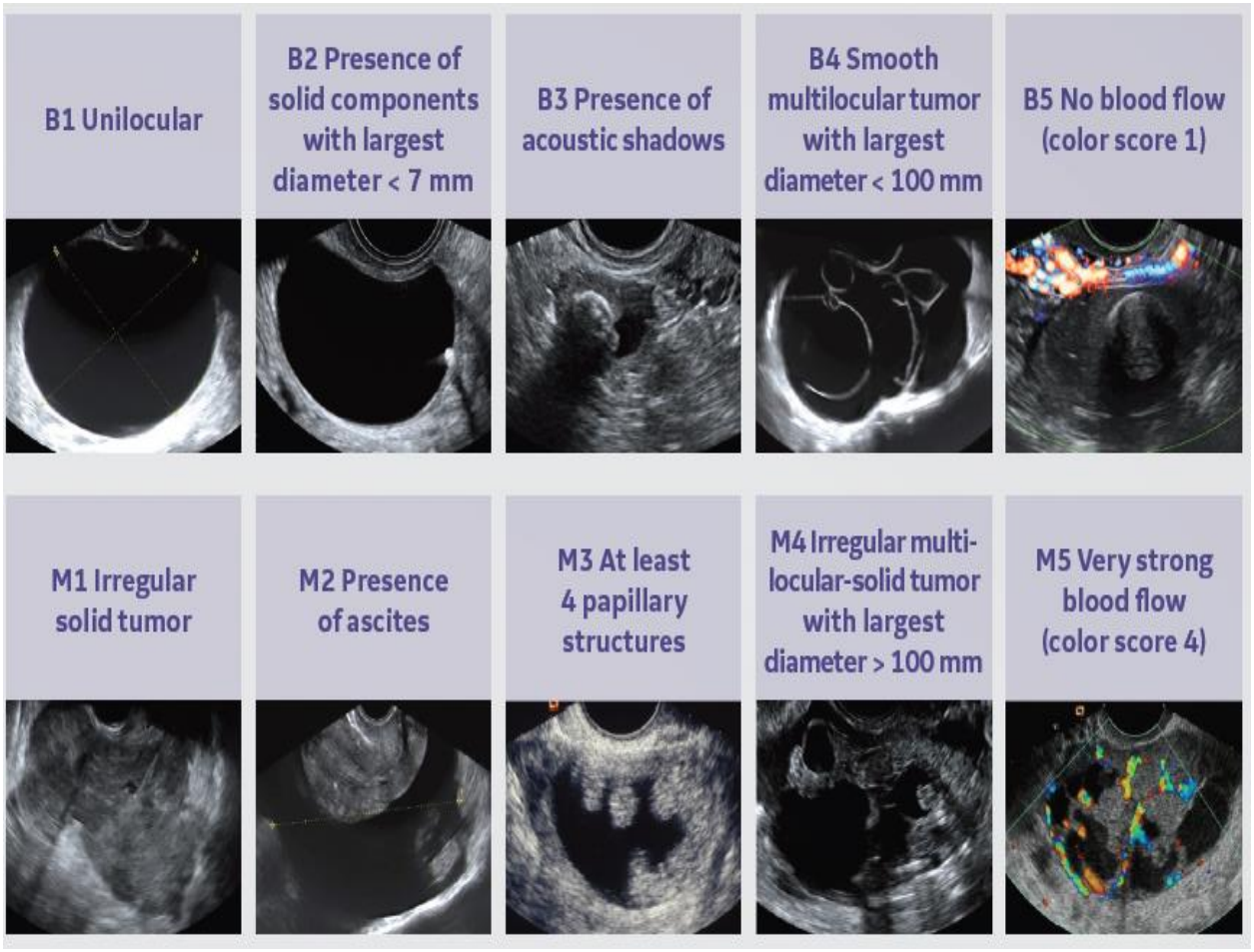

Figure 5: Grades of B rules and M rules.

At term patient underwent elective cesarean section for twins on 29 March 2023 and delivered healthy female baby 2.450 kg and female baby of 2.250 kg with Apgar score 7 and 8 respectively.

Luteoma was noted at the time of caesarean section as shown in Figure 4.

Androgen levels came to baseline by 4 weeks postpartum. Luteoma of pregnancy resolved by 3 months after delivery and hirsutism got resolved after 4 months of delivery.

DISCUSSION

Polycystic ovary syndrome, advanced maternal age and multiple pregnancies are risk factors for luteoma in pregnancy. Women who have already had a luteoma during a previous pregnancy have a higher risk of having luteoma in future pregnancy.[5]

Typical ultrasonographic characteristics include a solid, complex-appearing unilateral or bilateral mass with cystic features that correspond to areas of hemorrhage. It is usually not possible to differentiate luteomas from other solid ovarian neoplasms, such as luteinized thecoma, granulose cell tumour, or Leydig cell tumour, based on ultrasound characteristics alone.[6] Human chorionic gonadotropin (HCG) is responsible from the increased and uncontrolled cell proliferation on ovarian stroma and this pathophysiological mechanism may be the underlying reason for pregnancy luteomas.[5,7] Majority of these rare lesions are detected incidentally during cesarean section without any complaints.[5,7]

In a study pregnancy luteomas were detected during cesearean section. Luteomas on gross examination were found to be solid, soft, tan, or flesh colored, with hemorrhagic foci.

Microscopically, luteomas were seen as sharply circumscribed nodules composed of polygonal cells arranged in sheets, cords, or small clusters or form follicles containing colloid-like material.

The cytoplasm is abundant eosinophilic and finely granular. The nuclei may be slightly pleiomorphic. In 25% of the cases, luteomas are hormonally active leading to secretion of androgens causing maternal hirsutism and virilization. Table 1 shows differential diagnosis of ovarian tumors.

In a study where 18 cases of pregnancy luteomas were studied in multiple pregnancies the gross, immune histochemical and reticular fiber stain may help diagnose the disease.[8] Luteoma of pregnancy is often mistaken for having malignant potential and sometimes resulted in surgical removal during 1st trimester.[9] So, detailed work-up of every adnexal mass during pregnancy is important. Pregnancy luteoma can be differentiated from ovarian malignancy by "International Ovarian Tumor Analysis (IOTA) simple rules" on ultrasonography which was based on tumor size, architecture, wall contour, presence of ascites/ acoustic shadows and doppler flow velocities as shown in Figure 5.[10] Also, tumor markers (CA-125, Alfa fetoprotein, HE-4), hormonal assay (which includes testosterone levels, androstenedione and dihydro-testosterone levels, sex hormone binding globulin/ HSBG levels) and MRI if required can be done for differentiation. IOTA simple rules are classified as benign (B rules) and malignant (M rules).

Although luteomas regress after delivery, they may recur in subsequent pregnancies.[11] Pregnancy luteomas may result in maternal virilization, but usually the female fetus is not affected. This is presumably because of the protective role of the placenta and its high capacity to convert androgens and androgen-like steroids to estrogens.[12] With high clinical suspicion for pregnancy luteoma, clinical monitoring and postpartum radiologic follow-up may be an appropriate management strategy to avoid unnecessary surgery.

However, in some cases with atypical presentation or with complications from the mass, surgical intervention may be necessary for diagnostic or management purposes. Patients who present in the first half of pregnancy generally have more severe symptoms and are more likely to require surgical intervention for management of mass effect. When there is a high clinical suspicion for pregnancy luteoma, conservative management is appropriate since these tumors will usually regress spontaneously.[7] Antenatal accurate diagnosis is challenging but extremely important in order to optimize the obstetrical management of the patient and to improve maternal and fetal outcome.[13]

CONCLUSION

Luteoma of pregnancy are rare and mostly resolves spontaneously, so treated conservatively. Surgical intervention is reserved if suspicion for malignancy/ symptomatic patients. Detailed and judicious work-up of every adnexal mass in pregnancy should be done by ultrasonography, tumor markers, hormonal assay for differentiating luteoma of pregnancy from malignancy and deciding appropriate line of management. Early detection of luteoma of pregnancy is necessary for prevention of preterm labour.

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