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## Case Report

# Massive collision tumor of ovary in pregnancy: rare successful perinatal outcome

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### ABSTRACT

A collision tumor is the co-existence of two adjoining but histologically distinct tumors without histological admixture in the same tissue or organ. The most common histological combination of collision tumor in the ovary is the coexistence of mature teratoma with mucinous cystadenomas. The incidence of adnexal mass in pregnancy is 0.05-2.4% of which 94-99% are benign. The 1-6% maybe malignant requiring more extensive surgical procedures that increases likelihood of pregnancy loss. Ovarian tumors first diagnosed during pregnancy often present a challenge for diagnosis and management. Hence accurate knowledge of tumor characteristics, especially the ultrasound appearance and gestational age at diagnosis, are the key prerequisites for establishing the most effective plan of management for the pregnant women. To the best of our knowledge this report of huge collision tumor with successful pregnancy outcome is first of its kind.

**Keywords:** Collision tumor, Oophorectomy, Pregnancy, Term delivery

### INTRODUCTION

As is evident in the name, a collision tumor is the co-existence of two adjoining but histologically distinct tumors without histological intermixing. Although rare in ovary, commonest histological combination is the coexistence of mature teratoma with mucinous cystadenomas. More commonly described in esophagus, liver, kidneys.

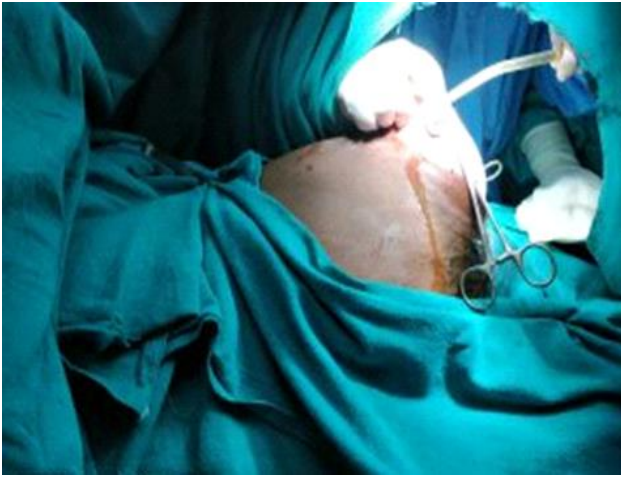
As the incidence of adnexal mass in pregnancy is low at 0.05-2.4%, every obstetrician may not come across such clinical dilemmas. Most adnexal masses (94-99%) are benign and only 1-6% maybe malignant requiring more extensive surgical procedures that increases likelihood of pregnancy lost.<sup>1</sup> Ovarian tumors diagnosed first time during pregnancy often present a clinical challenge. The presenting symptoms, ultrasound tumor characteristics pointing to benign or malignant nature of mass and the gestational age at diagnosis, are the key issues for

establishing the most effective plan of management in pregnancy.

This case highlights a huge (32 weeks) collision tumor presenting in early pregnancy, managed surgically at 16 weeks and with uneventful continuation of pregnancy and delivery of a live male baby at full term gestation. To the best of our knowledge this report of huge collision tumor with operative in successful pregnancy outcome is first of its kind.

### CASE REPORT

A 30-year fifth gravida having four living children, presented with one and a half months amenorrhea and hugely distended abdomen. Her complaints were of right sided abdominal pain for 2 weeks with loss of appetite, difficulty in micturition and constipation. General examination revealed otherwise healthy but pale patient. On her abdominal examination an abdomino-pelvic, cystic non tender lump of 32 weeks size was found (Figure 1).



**Figure 1: A 32 week hugely distended abdomen.**

Pregnancy was confirmed by a urine pregnancy test and ultrasonography showed a 10 weeks single live intra-uterine fetus. There was a large right adnexal complex mass of 30×23 cm, having small hyperechoic mural nodule with large multi-loculated cysts, wall was thin, no papillary projections and no increase in vascularity was appreciated on ultrasonography. Left ovary was also bulky measuring 5×4 cm. Other abdominal organs, including the liver, pancreas, kidneys, adrenals, and GI tract were reported as normal. There was no fluid in peritoneal cavity. Serum ovarian cancer markers CA 125 level (52.9 U/mL) beta HCG >50000 mIU/ml, alfa feto protein (18.2 ng/ml) and sonography pointed to benign ovarian tumor. She was explained the nature of mass and since she was having distress she was counselled for surgery. She was planned for surgery at 14-16 weeks of gestation, was prescribed progesterone and folic acid till 16 weeks.

She was kept under close scrutiny for next 6 weeks with progesterone supplementation and underwent exploratory laparotomy in second trimester (at 16 weeks) under spinal anaesthesia. Midline longitudinal incision at laparotomy revealed smooth shiny pale right sided ovarian cyst, which was drained through a small incision on its capsule to limit abdominal incision length (Figure 2). Minimum gentle handling of the gravid uterus was done.



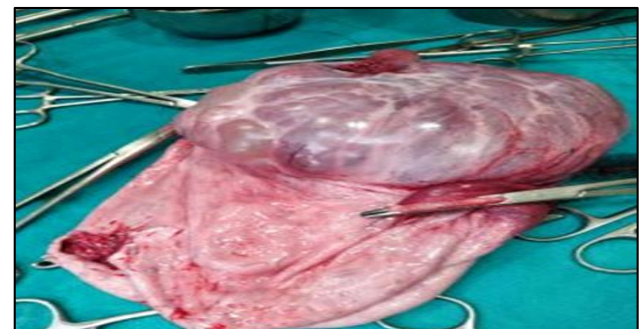
**Figure 2: Midline longitudinal infraumbilical incision and suction of ovarian cyst contents.**

Findings included a massive right ovarian cyst replacing the entire right ovary with no normal residual ovarian stroma (30×15×10 cm). The cyst was twisted twice on its pedicle (Figure 3).



**Figure 3: Exteriorized gravid 16 weeks uterus and bilateral adnexa. Right huge ovarian cyst with torsion in pedicle.**

Upper portion of cyst was 20×15×8 cm, 5.4 liters of dark colored serous fluid was aspirated from it (Figure 4 and 5).



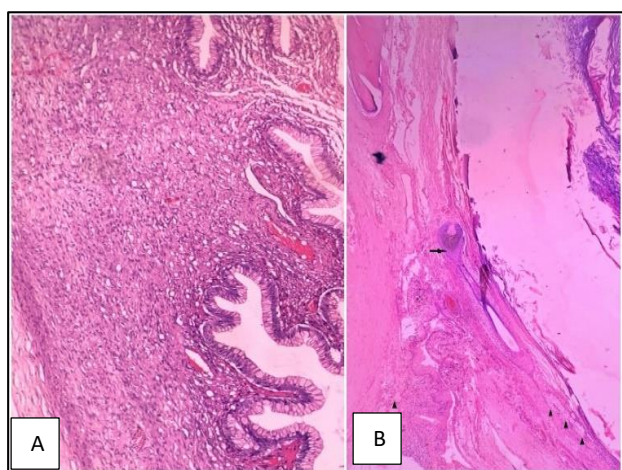
**Figure 4: Upper part of shows multiloculated cyst with a dermoid component, lower part if pic shows one large cyst which contained 5 l fluid.**



**Figure 5: Litres of hemorrhagic sero-mucinous fluid suctioned from cyst.**

Lower cyst was approximately 10×10 cm which contained seromucinous fluid and was multilobulated. One of the daughter cysts (4×4 cm) contained pultaceous material with tufts of hair and a nodule of 1X1cm, clearly a dermoid cyst. Left ovary contained a simple cyst of 4×3 cm. Right salphingo-oophorectomy was done with left cystectomy. Peritoneal cavity was systematically explored but no metastatic deposits were present. Post operatively patient recovered uneventfully, was prescribed progesterone supplements for 4 weeks and discharged with advice of close follow up.

Histopathology reported mucinous cystadenoma (irregular infolded lining epithelium composed of non-ciliated tall columnar epithelium containing cytoplasmic vacuoles) and a single mature teratoma (intraluminal wet keratin and heterologous elements in the fibro collagenous cyst wall including bone, hair follicle, sebaceous and pyloric type glands) also present in the same ovary (Figure 6).



**Figure 6 (A and B): Section from cystic tumour area showing cyst wall formed by ovarian stroma and lined by irregular infolded lining epithelium composed of non-ciliated tall columnar epithelium containing cytoplasmic vacuoles. Other area from the same ovary showed a cyst predominantly devoid of lining epithelium with intraluminal wet keratin (right side margin) and heterologous elements in the fibrocollagenous cyst wall including bone (upper left corner), hair follicle (arrow), sebaceous and pyloric type glands (arrowheads), (H and E; 200x magnification).**

The right fallopian tube was unremarkable. There was no capsular breach or lympho-vascular or perineural invasion seen. Peritoneal lavage was negative for malignant cells and positive for reactive mesothelial cells.

The patient thereafter had uneventful post-operative recovery. Further antenatal period was unremarkable in the follow up. She delivered a liveborn healthy male baby at 39 weeks gestation of birth weight 2900 gm.

## DISCUSSION

Collision tumors of the ovary are rare phenomenon. Meyer in 1919 defined it as the accidental meeting and intermingling of two independent tumours, which itself is quite a rare occurrence. They have been described for esophagus, liver, kidneys, thyroid, skin but very few cases have been reported in the ovary.<sup>2</sup> Occurrence of ovarian collision tumors in pregnancy becomes even rarer. The most common combination of tumors in ovary are a mature teratoma and a mucinous cystadenoma which was also present in this case.<sup>3</sup> Other reported histologic combinations have been of teratoma with granulosa cell, cystadenocarcinoma and sarcoma.<sup>4</sup>

This case highlights that even huge 32 weeks benign masses with pregnancy can be managed surgically in second trimester with uneventful continuation of pregnancy and normal delivery of healthy baby provided no operative complications occur. Another case reported of collision tumor in pregnancy had an acute abdominal presentation at 23 weeks gestation for which exploratory laparotomy was done but pregnancy outcome was not reported.<sup>5</sup> A 12 weeks pregnancy with collision tumor of right ovary having hemorrhagic mucinous cystadenoma and dermoid has been reported. The patient also had a dermoid cyst in the other ovary but with unknown outcome.<sup>6</sup> Table 1 compares the presentations, histology and pregnancy outcomes of few reported cases of ovarian collision tumors in pregnancy.

Various hypothesis which are postulated for the origin of collision tumors are the accidental occurrence of the two primary tumors. Or the primary tumors may have a common origin from same stem cell, or a similar carcinogen inducing both tumors simultaneously. There might be role of a favorable micro-environment like micro-angiogenesis and inflammation by one tumor providing for the growth of the second tumor, but all are unproven.<sup>7</sup> As is well known, benign cystic teratomas mostly occur in young female patients between 20-30 years of age being composed of derivative tissues of all three germ cell lines of ectoderm, mesoderm, endoderm. The mucinous cystadenomas are benign epithelial ovarian tumors which are multiloculated cysts, large and lined by endocervical epithelium producing mucin. Ultrastructure studies and mucin histochemical studies have supported the surface epithelial metaplasia theory.<sup>8</sup> Some proposals suggest their origin from metaplasia of surface epithelium or a teratomatous origin.

Homozygosity of chromosomal polymorphism have been seen in Molecular findings and they point towards germ cell origin of the ovarian mucinous cystadenomas associated with mature cystic teratomas.<sup>9</sup> Some other studies have suggested that these mucinous tumors have developed from pre-existing mature cystic teratomas in the ovary.<sup>10</sup>

**Table 1: Review of reported cases of ovarian collision tumor in pregnancy with outcomes.**

Study	Presentation	Histopathology	Pregnancy outcome
Aydin et al, 2019 <sup>5</sup>	23 weeks of gestation, acute abdomen	Mature cystic teratoma in collision with mucinous cystadenoma	Not reported
Shopov et al, 2019 <sup>7</sup>	38 weeks, in labour	Collision tumor of mature teratoma with a serous cystadenoma	Caesarean section with oophorectomy.
Suárez et al, 2019 <sup>8</sup>	Term with pain abdomen	Mucinous cystadenoma associated with a benign cystic teratoma of the ovary.	Caesarean section with oophorectomy.
Thakur et al, 2014 <sup>6</sup>	12 weeks pregnancy pain abdomen	Hemorrhagic mucinous cystadenoma and benign cystic teratoma along with dermoid cyst in left ovary	Not reported
<b>Present case</b>	8 weeks with hugely distended abdomen	Mucinous cystadenoma with mature teratoma, contralateral serous cystadenoma	Operated for ovarian pathology 16 weeks gestation, normal delivery at full term gestation

Widespread use of ultrasound in early pregnancy has led to the detection of asymptomatic incidental adnexal masses more frequently. Characterization of an ovarian mass with irregular solid mass, septations, papillary projections, ascites and strong blood flow helps predict malignancy. Dermoids are unilocular cysts with characteristic mixed echogenicity and acoustic shadows on sonography.<sup>11</sup> The possibility of collision ovarian tumor should be considered by sonologists when some additional findings other than typical imaging pointers of dermoid cyst are present.

Although many tumor markers are used to diagnose ovarian cancers in non-pregnant state, their interpretation is difficult as their levels are usually elevated in pregnancy.

Management can be conservative if it is certain that ovarian mass is benign, also 70% of adnexal masses detected in first trimester resolves by 2<sup>nd</sup> trimester. Persistent ovarian tumors and those >5 cm should raise suspicion for malignancy and surgical treatment is warranted. Complications of ovarian tumor in pregnancy like torsion, rupture or hemorrhage should also be considerations for surgery at the opportune time to have a favorable pregnancy outcome. Ovarian tumors particularly dermoids in pregnancy may cause torsion which occurs in 5% cases but out of them 60% occurs in second trimester. Second trimester is also the most favorable time for surgery as both the mother and fetal prognosis is good.

The histopathology should be evaluated by a specialized gynecology pathologist skilled in findings of ovarian tumor in pregnancy who has been communicated prior about presence of collision tumor if preoperatively suspected on ultrasonography. It is essential to exclude the possibility of malignancy in ovarian collision tumors to arrive at the appropriate histopathological diagnosis.

As this patient had benign components of both histological

types of tumors; hence no further treatment was required. The more aggressive partner of the collision tumor decides further line of management for the patient.

## CONCLUSION

Dermoid cysts are most common benign adnexal masses in pregnancy. The possibility of a second tumor (collision tumor) should be kept in mind and malignancy excluded, especially in larger masses. The most commonly partnered is mucinous cystadenoma. Symptomatic and suspicious collision tumor should be operated in second trimester of pregnancy to achieve best pregnancy outcome.

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