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CASE REPORT

COEXISTENCE OF STUMP APPENDICITIS AND OVARIAN FIBRO-THECOMA IN A YOUNG WOMEN WITH RECURRENT RIGHT ILIAC FOSSA PAIN DURING PREGNANCY, A RARE CASE REPORT

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ABSTRACT

Ovarian fibro-thecoma is a rare, benign, sex cord-stromal neoplasm, with a typically unilateral location in the ovary, characterized by mixed features of both fibroma and thecoma. It is a tumor of gonadal stromal cell origin accounting for 3-4% of all ovarian tumors. We describe a case of fibro-thecoma in a 27- year women during pregnancy associated with fever and vomiting who had previous history of laparotomy for appendicular abscess. She presented to us during pregnancy with this complain for which she was managed conservatively. She did not maintain her follow up regularly at our hospital and visited again after delivery of her baby with a still birth outcome with a newly diagnosed complex bilateral ovarian cyst demonstrated on ultrasound and computed tomography showed inflammatory right sided tubo-ovarian mass along with inflammatory thickening of ileum in right lower quadrant adjacent to right ovarian mass lesion. The patient underwent laparotomy for this with the removal of mass along with the removal of appendicular stump for appendicular stump appendicitis which was diagnosed intraoperatively. The finding from histopathological examination of the mass was consistent with the diagnosis of fibro-thecoma.

Keywords: *Fibro-thecoma, Ovarian neoplasm, Pregnancy, Sex cord stromal tumors, Stump Appendicitis.*

INTRODUCTION

Stump appendicitis is one of the rare delayed complications after appendectomy with reported incidence of 1 in 50,000 cases.[1] Residual tissue left after an initial appendectomy risks the development of stump appendicitis. Stump appendicitis can be seen in pregnant women and few cases have been reported showing stump appendicitis leading to chorioamnionitis in pregnant women.[2]

The Sex cord-stromal tumors are a distinct group of ovarian tumors representing about 1% - 4.7% of all ovarian neoplasms.[3] Most of them are unilateral, however bilateral cases may occur especially in patient with Gorlin syndrome.[4-6]

In the presented case, both of these rare disease stump appendicitis and ovarian fibro-thecoma co-existed in a young pregnant woman. Due to pregnancy and limitations of radiological investigations there was lots of dilemmas for the diagnosis and management of the patient. The objective of this case is to highlight the diagnostic evaluation and dilemmas faced during surgical management of these entities with a brief review of literature.

CASE HISTORY

We present a rare case of a 27 year old pregnant patient from low socio-economic background with recurrent right iliac fossa pain during her late second trimester and late third trimester with previous surgical history of laparotomy for appendicular abscess. She was Gravida 3 parity 2 living 2 with previous uneventful normal vaginal deliveries and presented to us during her late second trimester of pregnancy with complain of pain over right iliac fossa, insidious in onset, dull aching in nature, intermittent non-radiating without any aggravating factor which was associated with dyspepsia, constipation and vomiting. She had also presence of low- grade fever. Her blood, urine and radiological (Ultrasonography) investigation were done which showed normal findings

so the patient was managed conservatively. Her Fetal parameters were within normal range and was growing appropriately according to the period of gestation. She was discharged on fifth day when she was relieved from her symptoms with danger signs explained and was told to follow-up after 2 weeks. She did not maintain her follow up.

The patient again visited our obstetrical emergency in late third trimester with the similar complains. She was again evaluated and her Ultrasonography finding showed bilateral adenexal cyst. USG pelvis showed complex Bilateral ovarian cyst with internal echoes and septations with differential diagnosis of hemorrhagic cyst/ Endometrioma.

She was perceiving adequate fetal movement and baby was growing according to the gestational age. There was no history of per vaginal discharge/leakage/bleeding. Patient was advised for admission but they refused. After 7 days of discharge from hospital she delivered at nearby Primary Health Care center with a still birth outcome.

Her puerperium was apparently uneventful and after one month of delivery she felt a lump over right iliac fossa associated with pain which was initially small and later gradually increased in size. There was no history of decrease appetite, weight loss, dyspareunia, increased frequency or urgency of urine, night sweats, per vaginal discharge, cough and fever. She visited our hospital with this complain along with Computed Tomography abdomen and pelvis report which was done at some private hospital which showed the finding of Complex, multiloculated, multiseptated Right ovarian cyst with thickened septations with a differential diagnosis of Ovarian Malignancy and Inflammatory Tubo-ovarian mass along with Circumferential symmetrical thickening of ileum in Right lower Quadrant adjacent to Right Ovarian Mass lesion suggestive of Inflammatory thickening. Small left ovarian cyst with thin septation intermediate cystic lesion with enlarged mesenteric and right para-caval lymph node. Her tumor Markers were sent which were Lactate Dehydrogenase (LDH): 161, Cancer Antigen (CA-125): 2.5, Carcinoembryonic Antigen (CEA): 0.01.

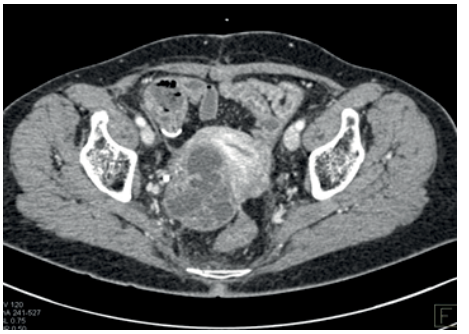

Fig 1: CT abdomen and pelvis showing right sided ovarian mass

As there was differential diagnosis of Ovarian Malignancy and inflammatory Right tubo-ovarian mass in the Computed Tomography scan report, patient and attendant were already counselled about the need of conservative surgery that is excision of the mass as well as chances and need of removal of uterus along with tubes and ovaries at some private hospital. However, they visited our hospital for opinion where we counselled them about the nature of the mass, need of frozen section biopsy and it's in availability at our hospital, risks and benefits of conservation of ovary, need of Hormonal Replacement Therapy along with the complications associated with extensive surgical procedure if needed.

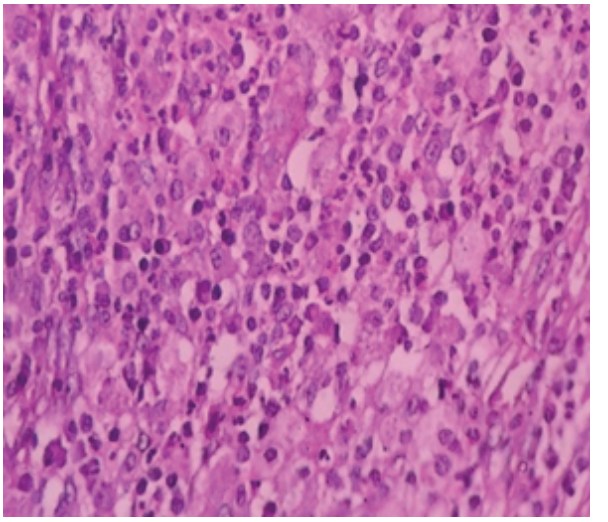

Fig 2:H&E Stain-High Power view of Fibro-thecoma (40X)

Patient underwent exploratory laparotomy proceeding to Total Abdominal Hysterectomy with bilateral Salpingo-ophorectomy along with removal of appendicular stump at our hospital with intraoperative finding of right sided tubo- ovarian mass measuring 5x6cm with dense adhesion of bowel loops with right sided tube and ovary and right lateral wall of uterus. Left sided simple ovarian cyst measuring 2x3cm with normal fallopian tube. Left sided Cyst contained smooth wall with clear fluid. 10 ml of pus was present in between intra bowel loops adhesions. This finding raised suspicion of some additional pathology so mobilization of caecum was done by the General Surgeons team. Appendicular stump of 1cm was present at the base of caecum at the postero medial aspect which showed sign of stump appendicitis along with small tiny perforation at the base of appendix. Patient's husband was again counselled explaining about the intraoperative finding and explained about conservation of contralateral ovary however he insisted on removal of uterus and ovaries and agreed on Hormonal Replacement Therapy.

Gross finding of Right Ovary includes dimension of 4.5 X 4 X 2.5 cm. Outer Surface is solid, firm, grey white and cut section shows solid homogeneous grey white surface with dark brown

Microscopic finding includes sections from the right ovary which shows spindle cells arranged in the fascicles. The cells are spindly elongated with tapering ends and have mild to moderate eosinophilic cytoplasm with elongated nucleus. Few of the cells are having cytoplasmic vacuoles and round to oval nucleus. Few foci of hemorrhages are also noted. There is no evidence of atypia. (Figure 2)

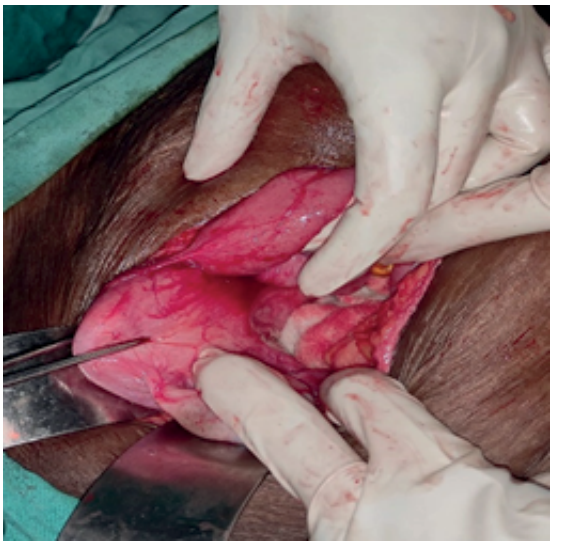

Fig 3: Right sided tube and ovary buried cyst the adhesions with bowel loops.

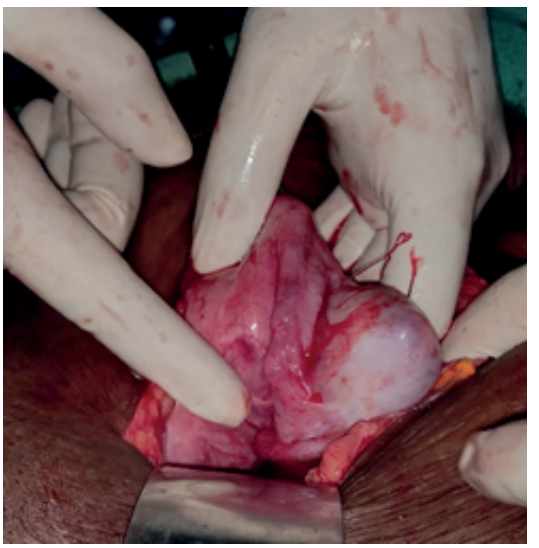

Fig 4: Left sided tube and ovarian cyst

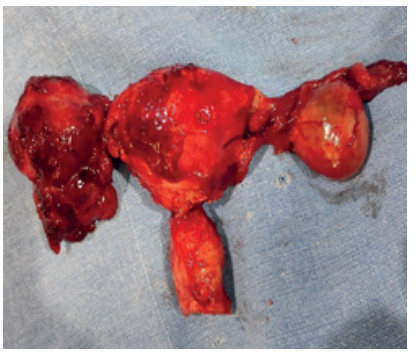

Fig 5: Specimen of uterus with bilateral tubes and ovaries.

DISCUSSION

This case is a challenging case creating a lot of dilemmas for diagnosis. The clinical features with which the patient presented to us created confusion between real pathology and pregnancy symptoms. Moreover, Ultrasonography done during late second trimester failed to give definitive diagnosis because it has some limitation due to increased uterine volume making evaluation difficult. First presentation of the patient at our hospital was in late second trimester. Furthermore, fluxometric parameters may be difficult to appreciate in pregnancy, both by increased flow velocity and lower vascular resistance found at pregnancy and by the significant overlap in fluxometric pattern between benign and malignant masses with an estimated false positive rate of 48-49%.[7,8] As in our case Ultrasonography was done when the patient was gravid with these limitations which failed to give definitive diagnosis.

Ovarian fibro-thecoma is often difficult to diagnose pre-operatively on clinical and radio imaging.[9,10] As in our case the ultrasonography findings were suggestive of Hemorrhagic cyst or Endometrioma and CT scan finding were suggestive of ovarian malignancy or Inflammatory Right tubo-ovarian mass. In 79% of the cases the tumor appears as a solid mass with delayed accumulation of contrast medium, while in 21% of the cases the tumor is partly or mainly cystic thus making differential diagnosis from other ovarian masses such as serous cyst adenofibromas or even malignant tumors difficult.[7] As in our case on CT diagnosis of Ovarian Malignancy or Tubo-ovarian abscess was made. MRI was not used as an imaging modality in this case which could have otherwise suggested Fibro-thecoma as a differential diagnosis and would have prevented an excessive surgical intervention especially if the patient is in younger age group.

In our case it was stump appendicitis which resulted in recurrent right iliac fossa pain in the patient and fibro-thecoma was sitting silently creating no harm to the patient. However preoperatively diagnosis was linked to ovarian pathology. The aggressive behavior of this benign ovarian fibro-thecoma is very rare. As in our case it was inert and all symptoms arise due to stump appendicitis. The tumor markers in our case were within normal range. Due to Appendectomy history, Stump appendicitis diagnosis was not kept in mind which resulted in increased morbidity. The circumferential symmetrical inflammatory thickening of ileum in right lower quadrant adjacent to right ovarian mass lesion in CT findings were suggestive of it in our case.

The purpose of a Frozen section biopsy is to provide with information about the microscopic amounts of tumor which cannot be seen with the naked eye and helps with a preliminary diagnosis of whether a suspicious mass is

benign or contains malignant cells. Due to lack of facility for this it was not done in our patient. As the patient had to underwent major surgery for the second time with in a period of ten years and their family was also complete, they were reluctant for conservative surgery.

CONCLUSION

In all patients presenting with recurrent right iliac fossa pain, stump appendicitis should be ruled out if the patient gives the history of appendectomy in past. Since Sex cord stromal tumors like Fibro-thecoma is benign and has less aggressive behavior, radiological investigations like MRI should be done to prevent extensive surgical procedure especially in young patients.

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