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Extra-gestational choriocarcinoma of colon - Diagnostic challenges and management issues

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ABSTRACT

Choriocarcinoma is an unfrequently encountered malignant tumour arising from the placenta in the females and the gonads in males. Extra-gonadal or non-gestational choriocarcinoma are rare tumors which arise from sites which include the mediastinum, lung, stomach, cervix, ureter and pancreas. Primary choriocarcinoma arising from the rectum or the colon is extremely rare to encounter with very few cases reported till date. It is usually associated with poor prognosis. We present a case of 29 year old third gravida with known case of rheumatic heart disease who was referred to our rural tertiary care centre with a diagnosis of probable ruptured ectopic pregnancy which turned out to be a case extra gestational choriocarcinoma of the colon upon investigations. The patient was started with chemotherapy however she succumbed due to pulmonary embolism secondary to her high risk pregnancy due to rheumatic heart disease. This case highlights that though extra gestational choriocarcinoma is rare to encounter it is important for the clinicians to be vigilant for the same as it is often a misdiagnosed condition specially in the rural peripheral centres where there is lack of resources and investigations to confirm the diagnosis.

Keywords: extra gestational choriocarcinoma, primary choriocarcinoma of colon, ruptured ectopic

1. INTRODUCTION

Extra-gestational Choriocarcinoma is a rare tumor and that from colon is much rarer, with only 19 cases reported till date (Boyce et al., 2020). Only four of these are pure primary Choriocarcinoma of the colon (Boyce et al., 2020). Existing information about the frequency, diagnosis and management of Primary choriocarcinoma of the colon is not well established (Iliev et al., 2017). Approach to diagnosis presents challenges especially if the clinical presentation and history is dubious (Mangla et al., 2017). We are reporting one such case wherein the woman was referred as ectopic pregnancy with severe

anemia and clinically debilitating condition. The 29-year-old, G3P1L1A1 reported to the emergency unit of a rural tertiary care center, with weakly positive pregnancy test done by referring center and accompanying pelvic ultrasound report suggesting the diagnosis of ruptured ectopic pregnancy. However, laparotomy did not reveal ectopic pregnancy, and uterus tubes and ovaries were normal. Suprapubic small transverse incision did not favour further exploration due to patients debilitating cardiovascular condition. Postoperative exponential rise of serum beta HCG warranted investigation by ultrasonography followed by CECT (abdomen+pelvis+thorax) which revealed large abdominal masses with mediastinal additional masses which were initially reported as Non-Hodgkin's Lymphoma by radiologist. Initial biopsy from the bowel mass as well as mediastinal mass revealed necrotic tissue, which further compounded the diagnostic dilemma, however further directed biopsy suggested histopathological diagnosis of choriocarcinoma of colon. We are discussing this interesting case for enhancing scientific knowledge and experiential learning. Team approach of gynaecologist, surgeon, interventional radiologist, cytopathologist, respiratory physician and oncologist is highlighted.

2. CASE REPORT

29 years old, third gravida with previous one living child and history of one abortion was referred to the emergency unit of a rural tertiary care center, with weakly positive pregnancy test done by referring center and accompanying pelvic ultrasound report suggesting the diagnosis of ruptured ectopic pregnancy. She was a diagnosed case of Rheumatic heart disease with severe anemia. On initial evaluation, her pulse was 110/ min, blood pressure was 100/60 mm Hg, severe pallor was present, generalized anasarca was present. On systemic examination cardiac murmur, bilateral crepitations in the lung, gross ascites along with severe tenderness in lower abdomen was present. On per vaginum examination tenderness was present in bilateral fornices. Uterine size could not be made out because of gross ascites. Her hemoglobin was 6 gm%. Exploratory laparotomy was done in view of suspected ruptured ectopic pregnancy with informed high risk consent. However, uterus, tubes and ovaries were normal on laparotomy and there was no hemoperitoneum. 500 ml of straw colored ascitic fluid was present in peritoneal cavity which was sent for cytology. Bowel mass covered by omentum on left side of the abdomen was visualised (Figure 1) along with lymph nodes in the right pelvic region embedded in the bowel (Figure 2).

Supra pubic small transverse incision did not favor further exploration due to patients debilitating cardiovascular condition. Anesthetist suggested prompt closure and reduced surgical time due to which presence of any other mass was missed. In view of positive pregnancy test, post operatively serum beta HCG was advised which came out to be 3653mIU/ml. It was repeated after 48 hours and rising trend was seen. Rise of serum beta HCG warranted investigation by ultrasonography followed by CECT (abdomen + pelvis + thorax) which revealed 2 large moderately enhancing soft tissue masses with adhered and embedded bowel loops in right lumbar region measuring 7.7 x 7 cm (left) and 8.8 x 5.5x x 6.5 cm (right) probably arising from large bowel (Figure 3). Multiple abdominal and pelvic lymph nodes were found to be enlarged almost as a chain attaining a size of up to 4x5 cm. Aneurismal dilatation of bowel was noted which prompted a radiological diagnosis of Non-Hodgkin's Lymphoma. Multiple rounded varied size peripherally enhancing conglomerate lesions with central areas of necrosis were seen within mediastinum suggesting metastatic lymphadenopathy. Bilateral Pleural effusion and pleural thickening was seen (Figure 4).

Histopathology of endometrial tissue suggested secretary endometrium and no evidence of pregnancy. Initial USG guided FNAC from the bowel mass as well as mediastinal mass revealed necrotic tissue, which further compounded the diagnostic dilemma. FNAC from retroperitoneal lymph node masses suggested mesothelial cell hyperplasia with infiltrate of polymorphous lymphocyte with no malignant cell and no granuloma. Pleural biopsy was planned, however patient collapsed during the procedure and had to be managed in intensive care. Further CT directed biopsy suggested histopathological diagnosis of choriocarcinoma of colon (Figure 5 and 6). Serum beta HCG value on day 5 was 12000 m IU/ ml in the rising trend. After discussing with oncologist, multidrug regimen chemotherapy (EMACO) etoposide, methotrexate, actinomycin D, cyclophosphamide, vincristine with guarded prognosis was planned. General condition was built up and chemotherapy was given under observation with first dose of Methotrexate. However, existing rheumatic valvular heart disease proved high risk for the patient and patient suffered fatal pulmonary embolism and could not survive beyond 15 days.

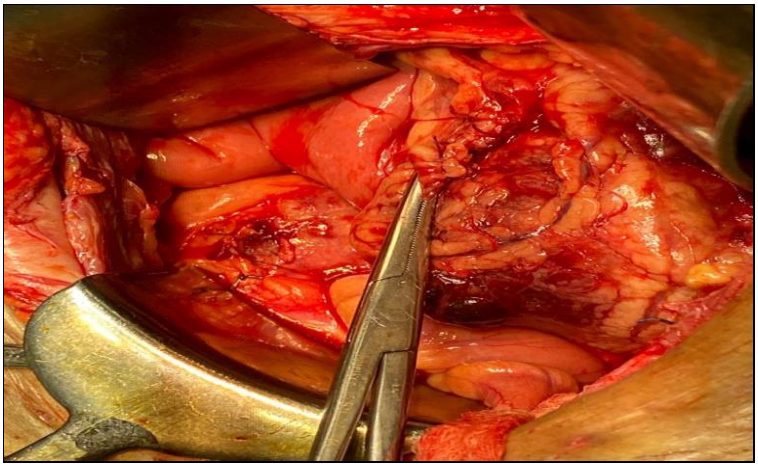

Figure 1 Bowel mass covered by omentum on left side of the abdomen

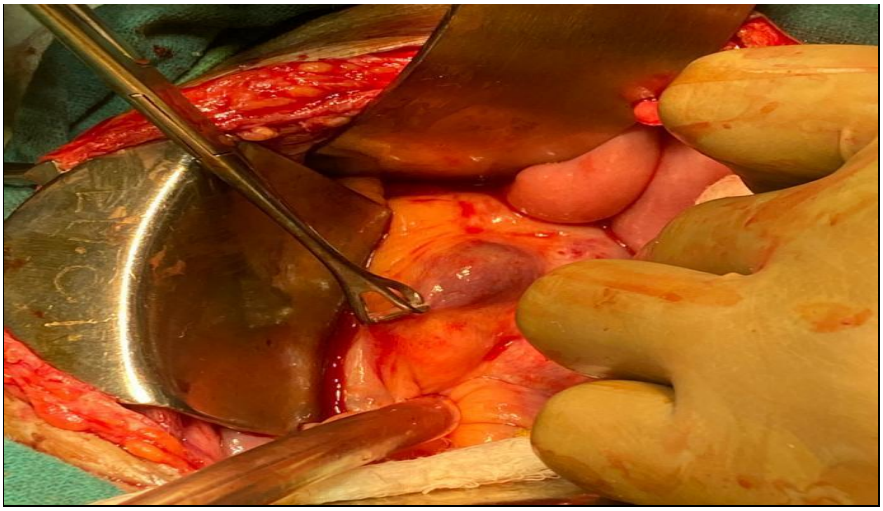

Figure 2 Lymph node in right pelvic region seen embedded in bowel

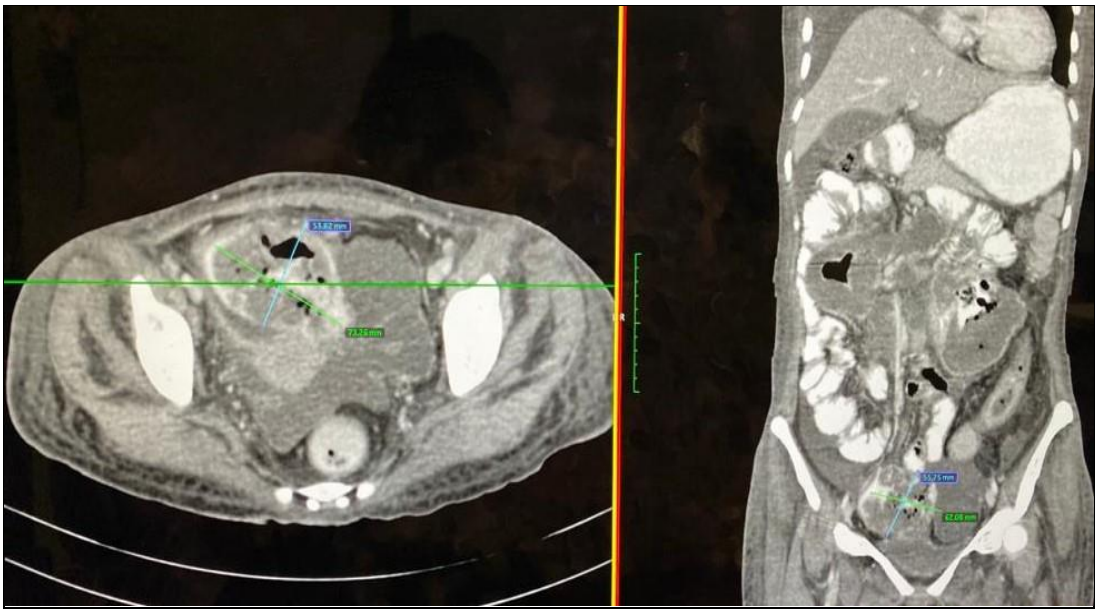

Figure 3 CECT abdomen multiple masses in abdomen

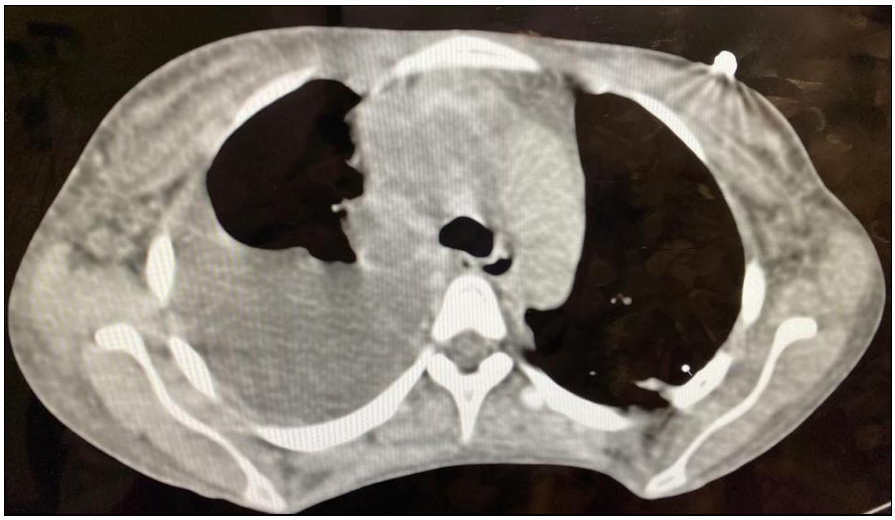

Figure 4 CECT thorax showing mediastinal mass

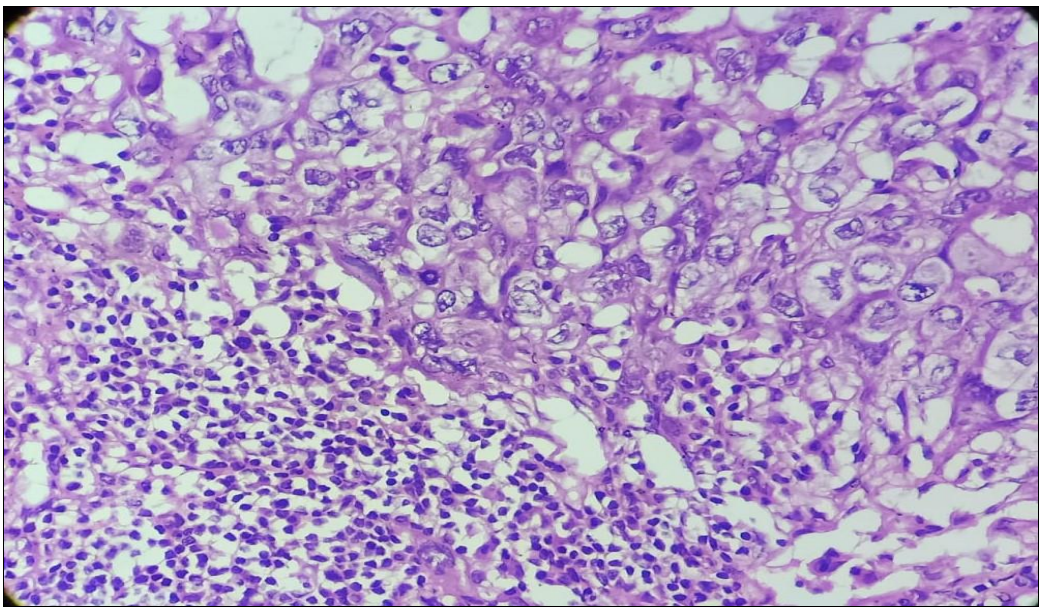

Figure 5 Photomicrograph showing malignant cells of colonic choriocarcinoma infiltrating the lymph node

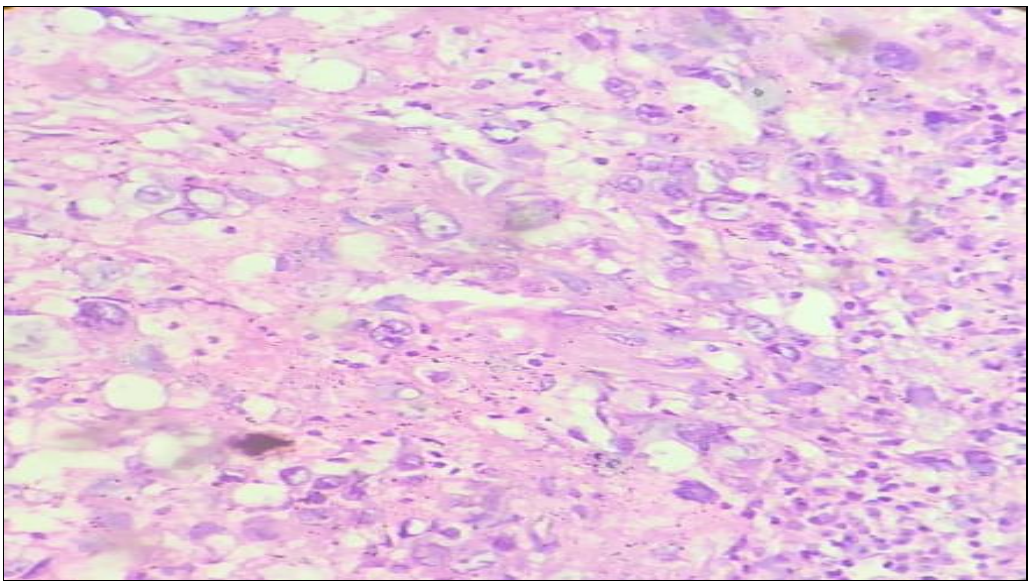

Figure 6 Photomicrograph shows malignant cells of colonic choriocarcinoma with necrosis (H and E slide, 40x)

3. DISCUSSION

Choriocarcinoma is a highly malignant tumor of trophoblastic cells that most often arises in the female genital tract, closely related to normal or ectopic pregnancy, molar pregnancy and abortion (Katke et al., 2015). In men, testes are the most common sites. Non gestational variety is known to occur in midline tissue and is found to affect the alimentary tract (Katke et al., 2015). Mori et al., (1982) reviewed 44 cases of gastric choriocarcinoma however, Primary Choriocarcinoma of colon (PCC) is very rare and has poor prognosis. There are huge diagnostic challenges for PCC if a woman presents with a positive pregnancy test and already has an ultrasound report indicating ruptured ectopic pregnancy as in the present case. Laparotomy was the natural choice, however a low transverse small incision, emergency hours, poor cardio respiratory status of the woman mandated a quick surgery and once uterus, tubes, and ovaries were found normal and hemoperitoneum was ruled out, it was advised by the anaesthetist staff to quickly finish the surgical closure. Any evidence of colonic mass was thus missed. A positive pregnancy test however directed further evaluation of covert gestation and dictated laboratory tests for serum beta HCG levels which were found very high and further rising after 48 hours.

Ultrasound and CECT both suggested large abdominal and pelvic masses as well as mediastinal spread. Such a clinical picture would definitely point towards a malignancy. However multiple attempts at histopathological diagnosis by directed biopsies only revealed inflammatory and necrotic tissue which did not match the clinical picture. Persistent effort was made to gather a larger chunk of tissue, and avoiding the central probably necrotic portion. That is when a diagnosis of PCC could be obtained. Such dilemma is more or less avoidable if primary surgery is able to pick up the colonic mass and histopathological diagnosis can be clinched. Jiang et al., (2013) also reported that preoperative diagnosis of PCC was poor as in his reported case intraoperative diagnosis was metastatic colonic cancer; even endoscopic biopsy could not pick up the lesion. However, excessive rise in beta HCG is the main clue which should definitely point towards chorionic tissue and in absence of gestational origin, extra-gestational choriocarcinoma is to be searched for.

The pathogenesis of primary extra-genital choriocarcinoma is controversial. Several hypotheses have been suggested but the retro-differentiation of pre-existing colonic carcinoma is the mostly accepted (Liu et al., 2001). Harada et al., (2012) proposed 4 possibilities; these elements are: (1) the result of dual differentiation of an immature malignant cell, (2) incidental coexistence of two different malignancy, (3) transformation of colonic carcinoma into choriocarcinoma, or (4) transformation of choriocarcinoma into colonic carcinoma, as there was coexistent colonic carcinoma and choriocarcinoma. In literature, most of the cases of colonic choriocarcinoma were associated with adenocarcinoma however, only 5 cases including ours were having the primary origin from colon (Julián et al., 2015). This phenomenon has been explained by Le et al., (2003) who proposed that a malignant change witnessed in the totipotent cells or the ectopic chorion results in choriocarcinoma. Reported cases of non-gestational choriocarcinoma were common in children and young adults before their 40s (Harada et al., 2012). It can be presented with a high beta HCG titer (Buza et al., 2014). Regional lymph nodes were involved in most of the cases and metastasis was reported in liver, lung and brain (Julián et al., 2015).

Most perished within 5 months of diagnosis (Harada et al., 2012). Present case had mediastinal spread. No relationship was found between the initial serum beta HCG level and prognosis (Mori et al., 1982). If the choriocarcinoma is in advanced stage with distant metastasis, chemotherapy will play an important role, because radiation and local treatment are thought to be ineffective however, there is no well-established protocol of chemotherapy (Julián et al., 2015). Most authors treated the co-existing colon carcinoma though it is reported that Chemotherapy for both the choriocarcinoma and the colonic adenocarcinoma is different. According to Harada et al., (2012) although there is no clear regimen of chemotherapy for primary colonic choriocarcinoma, combined therapy of MMC (mitomycin), MEA therapy (methotrexate, etoposide, actinomycin D), and UFT/leucovorin (5 Flurouracil) could be an effective treatment option.

However, overall prognosis is judged to be poor because its biological characteristics strongly favor generalized metastasis late diagnosis and distant liver and/or lung metastases (Katke et al., 2015). Present case had multiple metastasis and cardiovascular co morbidities which compounded the prognosis. Pulmonary embolism proved to be thee terminal event for our patient which in itself is a deadly complication with varied presentations ranging from benign symptoms (Kumar et al., 2009) to life threatening complications (Hulkoti et al., 2021) including seizures (Bagga et al., 2020) and cardiac arrest (Talwar et al., 2021). Choriocarcinoma is characterized by rapid proliferation with hematogenous or lymphogenous metastasis high clinical suspicion and early diagnosis is key player for survival.

4. CONCLUSION

Extra gestational choriocarcinoma has its own diagnostic challenges, especially if patient presents in a debilitating condition with acute obstetric emergency. However once gestation is ruled out rising beta HCG titers guide further evaluation of covert chorionic tissue. Primary choriocarcinoma of colon further eludes diagnosis and presents very late, thus prognosis is very poor in spite of advances in chemotherapy and surgical skill.

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Author Contributions

All the authors have contributed equally

Informed consent

Written & Oral informed consent was obtained from the patient's relative for this case report.

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Conflicts of interest

The authors declare that there are no conflicts of interests.

Data and materials availability

All data associated with this study are present in the paper.

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