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Rare presentation of aggressive choriocarcinoma in a 22-year-old female with splenic rupture after uncomplicated term pregnancy during extended puerperium: A case report

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Abstract
Choriocarcinoma (CC) is a rare, highly malignant pregnancy-related trophoblastic neoplasm that often presents with distant metastases. We present a rare case of a 22-year-old female who developed aggressive choriocarcinoma following an uncomplicated term pregnancy. The patient initially presented with severe headache, vomiting, and blurred vision. Imaging studies revealed venous infarct, thrombus in the superior sagittal sinus, and subdural hemorrhage. Subsequently, the patient developed sudden abdominal pain, with CT scans revealing splenic rupture, multiple organ lesions, and possible metastases. Despite an initial diagnosis of splenic rupture, a positive pregnancy test and high β-hCG levels led to the suspicion of choriocarcinoma. The patient was diagnosed with FIGO stage 4 choriocarcinoma, with splenic and brain metastasis. Histopathological examination confirmed metastatic deposits of choriocarcinoma in the spleen. The patient received combination chemotherapy (EMA-CO), but follow-up was lost due to treatment at another facility. This case highlights the aggressive nature of choriocarcinoma, its rare presentation, and the need for early suspicion and intervention in cases of unexplained post-partum complications.

Keywords: Choriocarcinoma, gestational trophoblastic neoplasia, splenic rupture, metastasis, case report

Introduction
Choriocarcinoma (CC) is a highly vascular and anaplastic neoplasm. It is part of a group of conditions wherein there is abnormal trophoblastic cell proliferation, known collectively as gestational trophoblastic neoplasias (GTN). It is a highly malignant pregnancy-related trophoblastic neoplasm and patients may manifest non-gynecological symptoms owing to distant metastases [1].

Gestational CC is a rare complication of previous molar and uncommonly non-molar gestation; it occurs mostly within one year of the antecedent pregnancy. Incidence of CC is approximately 0.6 of every 10000 normal gestations, 0.7 of every 10000 abortions, and in 250 of every 10000 molar pregnancy [2]. This tumour tends to invade vasculature and thus metastasis to the lung, vagina, brain, and liver. Therefore it can present with various organ symptoms [2].

Case Report
We report a case of 22-year-old woman who presented to medical emergency unit with symptoms of sudden, severe, throbbing headache associated with multiple episodes of non-bilious projectile vomiting and nausea for 14 days and sudden onset blurring of vision for 5 days.

She was para 2 live 2 with 11 week post-partum after full term vaginal delivery following uncomplicated pregnancy. She had initially normal lochia for 2 weeks and was then was in lactation amenorrhea since then.

On initial examination, there were no positive features except on fundoscopy which revealed left sided grade 2 papilledema and right sided grade 1 papilledema. Her baseline Complete blood count, renal and liver function test and coagulogram were normal. MRI revealed features suggestive of venous infarct involving right parieto occipital region with associated thrombus in superior sagittal sinus with Sub Dural haemorrhage. On brain and neck angiography there was a small spider like vessels the right occipital lobe s/o either venous angioma or Arterio-venous malformation.

On day 4 of her admission she developed sudden pain in left upper abdomen of the abdomen with abdominal distension. CT scan of abdomen reported bulky globular and large spleen 11.3 cm with hypodense areas in parenchyma predominantly non-enhancing with few foci of patchy enhancement. Lesions measured 65*50*55 mm and 26*35*41 mm. Similar lesions were noted in corticomedullary junction in bilateral kidneys, 37*30*47 mm in left mid and lower pole and 19*18*25 mm in right mid pole. Uterus was enlarged 87*56*59 mm with similar lesion measuring 54*50*42 mm in subserosal/intramural location on left side. Both ovaries were enlarged; left 34*47*33 mm and right 36*34*39 mm. There was significant free fluid in the abdomen. Limited section of thorax visible showed lesion in basal segment of right lower lobe of lung measuring 43*40*33 mm. Lesions were suggestive of probably multiple infarcts or Arterio-venous malformations or metastasis. Her Chest X-ray displayed cannon ball appearance of metastatic lesions in right lower and left upper lobe.

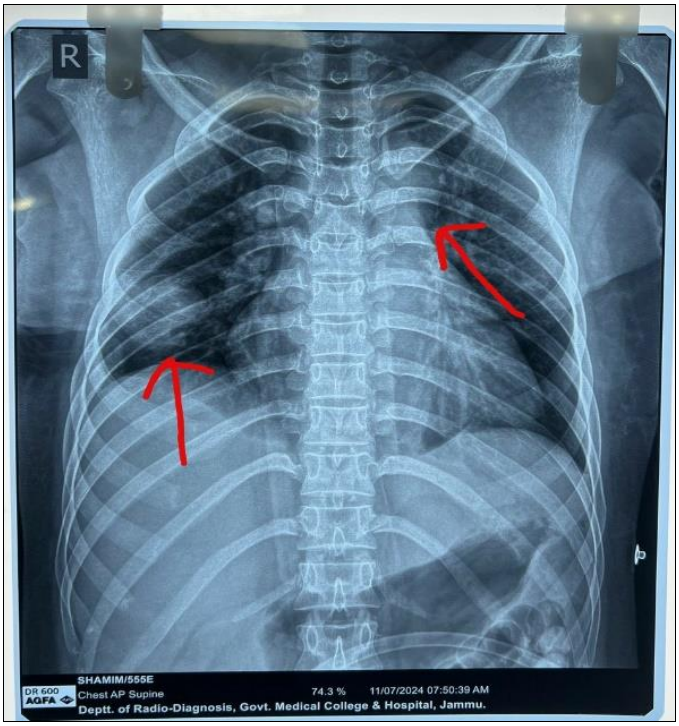

Fig 1: cannon ball lesions on X-ray chest PA view

She developed hypotension and tachycardia, abdominal paracentesis was performed which was hemorrhagic, thus decision of laparotomy was taken. A urine pregnancy test was performed which came out positive and sample was sent for serum beta-HCG. Consultation was then sought by the hospitals gynecology department.

Intraoperatively around 1.2 litre hemoperitoneum was present. Uterus was 10-12 weeks size with lesion present on left side of fundus measuring 3*3 cm, both ovaries were polycystic. There was no source of bleeding in the pelvis. On abdominal exploration it was found that there was splenic rupture. Splenic vessels ligated and spleen excised, other solid organs were inspected which were normal. Hemostasis was thus achieved. She received 2 units of packed cell and 2 units of FFPs.

Post operatively patient's B-hcg came out to be >1 lakh. A provisional diagnosis of choriocarcinoma was then made due to high suspicion and clinical picture, FIGO stage 4 with WHO prognostic score of 14 (<40 years old, 0, index pregnancy term 2, time since delivery < 4 month, 0, b-hCG >100000mIU/ml; 4, size of tumour > 50 mm, splenic metastasis 2, number of metastases 5-8; 2, site of metastasis-brain; 4) making this patient a high risk gestational trophoblastic neoplasia (GTN). She was started on treatment with combination chemotherapy agents; etoposide, methotrexate, actinomycin D, cyclophosphamide and vincristine (EMA-CO). Later on the histopathology of splenic tissue confirmed metastatic deposits of Choriocarcinoma. Consent was taken from the patient for publishing the case report ad images. Patient went to other institute later on for subsequent chemotherapy cycles, follow up could not be possible.

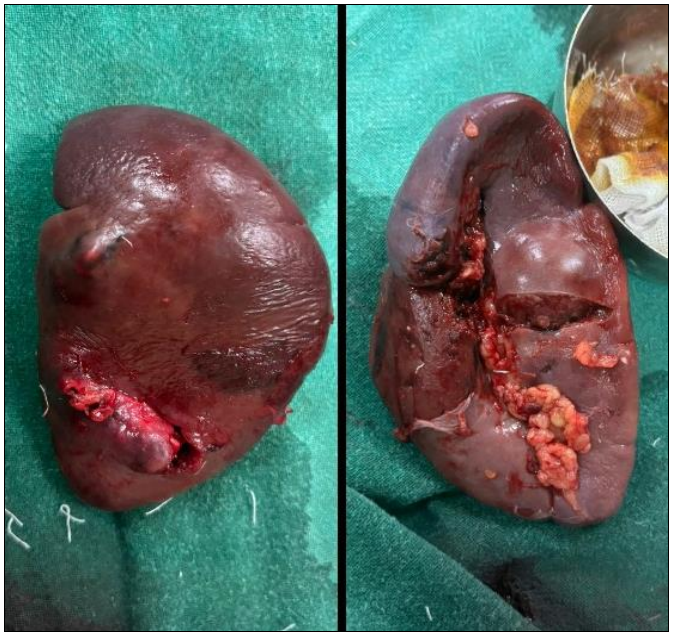

Fig 2: Excised ruptured spleen

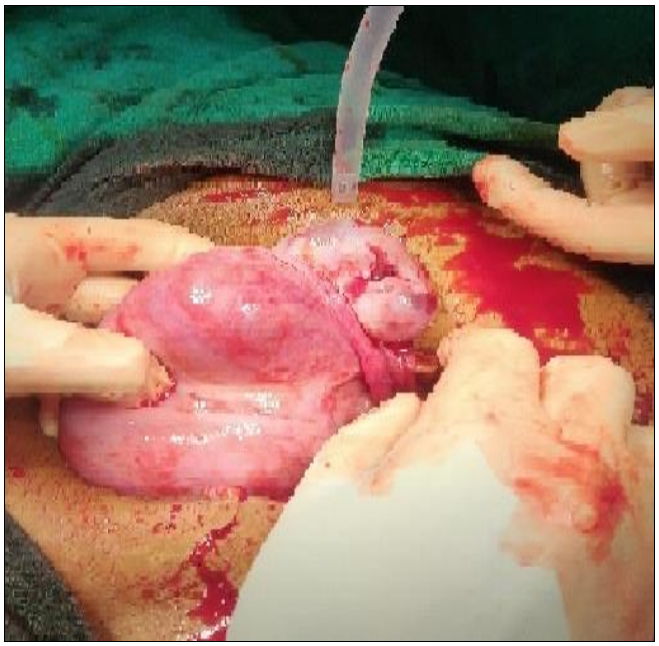

Fig 3: Lesion on anterior surface of uterus

Case Discussion

Choriocarcinoma after term pregnancy is a rare occurrence. It can present in women of any age. Nozad J, *et al*., reported choriocarcinoma in young female of 20 years of age, whereas Desai NR *et al*., presented a case of choriocarcinoma in 73 year old woman, 23 years after menopause [1, 3].

Since most of these cases occur following molar pregnancy, their detection rate is high due to early and serial serum beta HCG monitoring and availability of histopathological examination of molar specimen. Likewise even after ectopic pregnancy or miscarriage, beta-hcg is usually monitored till value of 0 and thus GTN is picked up early. Unfortunately, women with choriocarcinoma following a normal or preterm delivery potentially go undiagnosed as their B-hCG level is not routinely monitored following delivery and thus the disease can progress unchecked [4].

The clinical presentation of patient with CC differ depending on the site of metastasis causing delay in detection and diagnostic dilemma. Patients may present with cardiopulmonary complaints (20.66%), gastrointestinal bleeding (18.43%), or central nervous system symptoms (17.67%) [5]. The most common gynecological presentation of CC is abnormal uterine bleeding [4, 5]. This AUB was absent in our patient and thus prompt gynaecological consultation was not sought on admission.

Confirming the diagnosis with tissue histopathological examination (HPE) is a matter of debate and discussion, as the diagnosis is often evident from the history, the clinical picture, and the hCG levels. Like with our patient, taking into consideration the widespread metastasis and aggressive nature of tumour EMA-CO was started even before tissue HPE confirmation as she was only 11 weeks into puerperium and was in FIGO stage 4 GTN with score of 14. Patients with a FIGO score of <6 are considered low-risk and require single-agent chemotherapy such. In contrast, patients with a FIGO score of >7 are treated with multiple-agent regimens, most commonly EMA-CO. This approach has led to overall remission rates of approximately 98%-100% [6].

Conclusion

Identification of women with GTN is of utmost importance, it should be considered in the differential diagnosis of patients with unusual clinical presentation. In all such cases, serum B-hCG should be performed as part of initial workup for optimal and timely detection. Choriocarcinoma is a rare condition, and its diagnosis requires a very high level of suspicion by the treating clinician. Further investigations and studies are required to decide whether or not a tissue biopsy is needed before starting timely treatment in cases of GTN after term or preterm deliveries.

Conflict of Interest

Not available

Financial Support

Not available

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