Case Series

Gestational trophoblastic neoplasia with pulmonary embolism mimicking tuberculosis

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Abstract

Gestational trophoblastic neoplasia (GTN) comprises a group of human neoplastic diseases that derive from fetal trophoblastic tissues. They are proliferative as well as degenerative disorders of placental elements and include complete hydatidiform mole (CHM) or partial hydatidiform mole (PHM) (90%), invasive mole (IM) (5–8%), which could also be metastatic, villous, or villous choriocarcinoma (CC) (1–2%), and placental site trophoblastic tumor (PSTT) (1–2%). We present three cases of GTN, two mimicking tuberculosis radiologically, and all three are associated with pulmonary embolism.

Keywords: B-hCG level, chemotherapy, gestational trophoblastic neoplasia, pulmonary embolism, tuberculosis

Introduction

Gestational trophoblastic neoplasia (GTN) is classified histologically into three subgroups: choriocarcinoma destruens (invasive mole (IM)), choriocarcinoma (CC), and very rare placental site trophoblastic tumor (PSTT).[1] About 50% of all cases of GTN occur post-molar gestations, 25% after abortion or ectopic pregnancies, and 25% after term or preterm deliveries.[2]

CC and IM make up the vast majority of cases and produce B-hCG, and these are highly responsive to chemotherapy with an overall cure rate of 90%. B-hCG is used for the diagnosis, monitoring, treatment, and follow-up of these patients. However, the PSTT and epithelioid trophoblastic tumor (ETT) have low B-hCG levels; so, in these cases, surgery is used as first-line treatment as these are resistant to chemotherapy.[3] GTN mimics pulmonary tuberculosis radiologically, and it is always important to rule out GTN in young females especially.

Main Finding

We present cases of three young females presented within a year, non-smoker, and no other comorbidity with ongoing antitubercular drugs on the basis of chest X-ray and pleural fluid report.

First case—A 30-year-old woman, mother of a single live 5-month-old baby, presented to us with sudden onset of shortness of breath, hemoptysis (1/2 tsp in amount), and left side chest pain, on antitubercular treatment for 2 months. On examination, she had tachycardia with heart rate of 120/min and SpO2 level of 92% on room air; her blood investigation was within normal limit; she was evaluated further in view of drug resistance tuberculosis; however, all came out to be negative; on further evaluation, she was diagnosed as having pulmonary embolism; workup for the cause of embolism was initiated; on evaluating history, she came up with the history of occasional bleeding per vagina and a history of abortion

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around 2 years back; gynecological opinion was sought; and beta-human chorionic gonadotropin (B-hCG) levels were measured, which was >1 lakh, and further computed tomography (CT) abdomen conducted was suggestive of adnexal mass with normal scan for magnetic resonance imaging brain [Figures 1 and 2].

Second case—A 26-year-old woman, mother of a single live 2-year-old child, presented with sudden onset of shortness of breath, with no history of cough, chest pain, fever, and limb swelling. She was on antitubercular treatment for 3 months from outside in view of right tubercular pleural effusion [Figure 3]. On examination, SpO2 was 86% on room air and heart rate was 125/min in view of suspicious embolism; she underwent echocardiogram (echo) and CT pulmonary angiography and was diagnosed as extensive bilateral pulmonary embolism with infarct [Figure 4]; in view of this, she underwent embolectomy; and simultaneously workup for embolism was initiated and found to have B-hCG level >3 lakhs with history of spontaneous abortion 5 months back and was on ongoing contraceptive pills, though after embolectomy her saturation and condition improved. She underwent cerebrospinal fluid (CSF) examination too for B-hCG level, which was within normal limits.

Third case—A 34-year-old woman, mother of two alive children, the second child delivered 2 days before presenting to our hospital with a history of bleeding per vagina, underwent hysterectomy post-delivery and was then referred to us in view of worsening dyspnea; on examination, she looked pale and her saturation was 80% on room air, with tachycardia and tachypnea present, so there was high suspicious embolism; she underwent pulmonary angiography and revealed mass in the bilateral lung with cannonball appearance with embolism; on further investigation, she was found to have raised B-hCG levels; and she was planned for chemotherapy along with all symptomatic treatment; however, attendant refused for the same [Figure 5].

All three patients were initiated on heparin and later shifted to rivaroxaban and above 2 patients were initiated on chemotherapy etoposide, methotrexate, actinomycin D, cyclophosphamide, and vincristine (EMA/CO) regimen both of them are doing well and recovered from their illness and

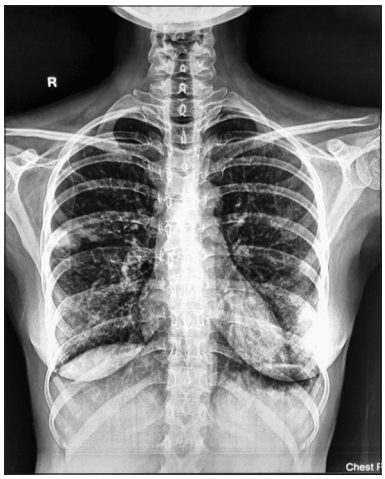

Figure 1: Chest X-ray—heterogeneous opacity bilateral lung fields

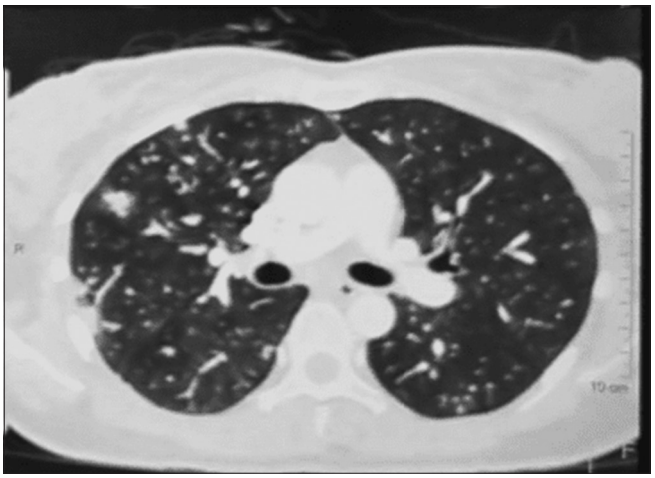

Figure 2: CT scan—shows multiple nodular pattern

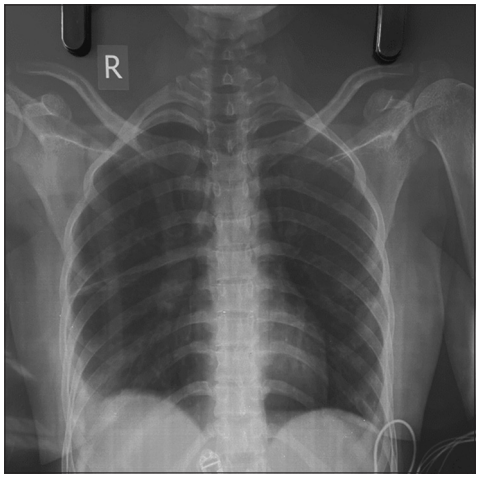

Figure 3: Chest X-ray—right pleural effusion

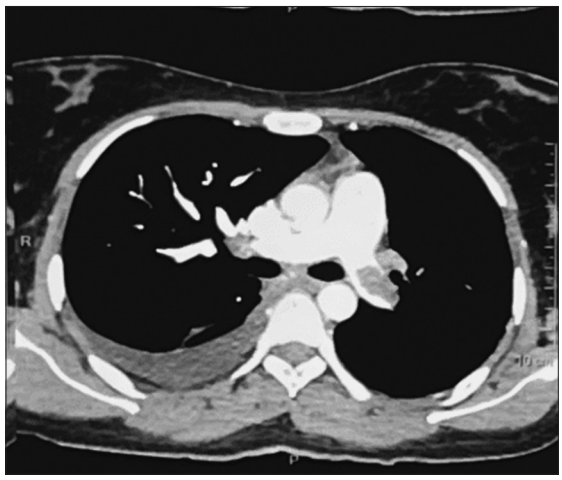

Figure 4: CT scan—right pleural effusion with bilateral pulmonary thrombosis

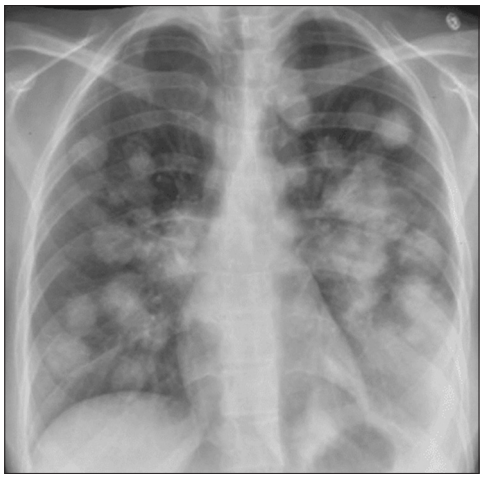

Figure 5: Chest X-ray—bilateral cannonball appearance

third one lost to follow-up, repeat pulmonary angiography revealed resolution of embolism.

Discussion

GTN is a rare, highly malignant neoplasm, which may occur during or following any type of pregnancy. It is caused by the abnormal proliferation of trophoblast cells in the placenta, which include benign mole and malignant trophoblast diseases, and has a low incidence ranging from 1 to 3/1000 pregnancies for hydatidiform mole (HM) to (1–9)/40,000 pregnancies for CC. The incidence of ectopic pregnancy (EP) is 1% to 2%.[3] There are four clinic pathological conditions—1) IM that follows either a complete hydatidiform mole (CHM) or partial hydatidiform mole (PHM), 2) CC, 3) PSTT, and 4) ETT. Each of these conditions can perforate the uterine wall, metastasize, and lead to death if left untreated.[1]

GTN develops in almost 15% of molar pregnancies.[4]

When the normal regulatory mechanism controlling the proliferation and invasion of trophoblastic tissue is lost, then GTN arises, and they have distinct tumor marker (B-hCG) originating from placental tissue and have varying tendencies toward local invasion and distant metastasis.[5]

In general, the sites of GTN metastases and the rate of spread to each site (among patients with metastases) include the following: pulmonary (80%), vagina (30%), central nervous system (10%), hepatic (10%), and other sites (kidney, gastrointestinal tract, and spleen).

The fundamental tool that can be considered for diagnosis of GTN is transvaginal ultrasound if a mole is invading local tissues, but there may be similar appearance on imaging studies. The most common is focal myometrial mass. The image may be hypo- or hyperechoic or complex and even multicystic.[6]

GTN has a varied presentation depending upon the antecedent pregnancy, extent of disease, and histopathology. GTN may follow a molar pregnancy (complete and partial) or any other pregnancy event (pregnancy loss, induced abortion, and preterm or term pregnancy). In our cases, all three patients were diagnosed after term pregnancy.

GTN following a term or preterm gestation may present with amenorrhea, but usually presents with abnormal uterine bleeding as in our case due to invasion of uterine tumor or bleeding from metastatic site. Bleeding from uterine perforation or metastatic lesion may result in abdominal pain, hemoptysis, or melena. Patients with central nervous system metastases often exhibit evidence of increased intracranial pressure from intracerebral hemorrhage, leading to headaches, dizziness, seizures, or hemiplegia. Patients who develop extensive pulmonary metastases may present with dyspnea, cough, or chest pain.

To evaluate lung metastasis, International Federation of Gynecology and obstetrics (FIGO) recommends a chest X-ray; however, 41% of patients with normal chest X-ray have lung metastasis on CT, and the presence of micro-metastasis does not seem to affect long-term survival.[7]

These are associated with pulmonary embolism either due to disease itself or seedling of tumor tissue in vessels giving the presentation of embolism. Metastatic GTN has mostly cannonball appearance radiologically; however, two cases of ours mimicked tuberculosis.

An elevated hCG is often the first evidence of possible GTN. A serum quantitative hCG should be drawn in all patients with prior molar pregnancy, non-molar pregnancy, no known history of pregnancy, and any female of reproductive age with evidence of metastatic disease without an obvious primary site of malignancy.

The scoring system for GTN is adopted by FIGO, and the treatment is based on the total score calculated. Patients can be treated with single-agent chemotherapy with either methotrexate or actinomycin D if it is a non-metastatic disease (stage I) and low-risk metastatic GTN (stages II and III, score <7); however, monitoring of drug resistance should be there as 5–15% may require multiagent chemotherapy. Etoposide, actinomycin D, methotrexate, vincristine, and cyclophosphamide (EMA/CO) or with cisplatin (EMA/CE) is usually first-line treatment in high-risk metastatic GTN, and our patient received both EMA/CO regimen and post-treatment follow-up of 6 months, and they are doing well.[8,9]

All patients must be encouraged to use effective contraception during the entire interval of monitoring oral contraceptive being safe.[10]

So, in young female patients presenting with abnormal per vaginal bleeding, radiologically mimicking tuberculosis with mostly a history of abortion, it is always must to rule out GTN, and not all patients require antitubercular treatment until proved to be tubercular.

Conclusion

All infiltrates on chest X-ray are not always tubercular, and it is always important to rule out GTN in young reproductive age-group females with pulmonary embolism.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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