

CHORIOCARCINOMA WITH METASTASES LEADING TO ARTERIOPORTAL FISTULA WITH PORTAL ANEURYSM

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ABSTRACT

Choriocarcinoma, a highly malignant neoplasm arising from syncytiotrophoblast, occurs as a rare condition in about 1 in 50,000 pregnancies. It tends to metastasize early, commonly to the lung, vulvovaginal region, brain, and liver.

A rare case is presented of a young female presenting with obstructive jaundice, vomiting, and diarrhea, that after clinical workup and CT scan, was traced to a large pseudoaneurysm in the pancreaticoduodenal region with possible intra-biliary rupture, causing mass effect on duodenum and ampulla resulting in obstructive jaundice. A large mass with a cystic component was also present in the uterus. A history of abortion two years back was obtained, and laboratory reports showed high β -HCG levels. Metastatic nodules were also present in the lungs and liver.

A diagnosis of choriocarcinoma with metastatic disease was made and the arterioportal fistula with portal aneurysm embolectomized surgically. She remained stable postoperatively and was further managed by the oncologist.

The authors declared no conflict of interest. All authors contributed substantially to the planning of research (SK, ZD), data collection (SK, ZS, SA), data analysis (SK, ZD) and write-up of the article (SK, ZD, ZS, SA), and agreed to be accountable for all aspects of the work.

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INTRODUCTION

Choriocarcinoma is a neoplasm of syncytiotrophoblastic origin and is the most malignant and aggressive neoplasm of all the gestational trophoblastic Diseases (GTD). It is very rare (1:50000 pregnancies) gynecological malignancy that can occur after molar pregnancy (50%), abortion (25%), term (20%) or ectopic pregnancy¹. Choriocarcinoma is associated with a high human gonadotrophin (HCG) level and its rapid hematogenous to multiple sites^{1,7,8,12,20,22,23,25}. The most common sites for metastases are lung and vulvovaginal region while the less common are brain and liver. Other sites for metastases are skin, gastrointestinal tract, kidney breast or bone but these are extremely rare. Although it can be effectively treated with chemotherapy but then occurrence of metastases put the person life at risk by developing life threatening hemorrhage due to necrosis caused by chemotherapy.

Interventional radiology (IR) is a fast-developing branch of radiology that has introduced new treatment modalities for internal bleeding and for the treatment of many other diseases.

In this article we present a rare case of a young lady diagnosed with choriocarcinoma that presented with jaundice.

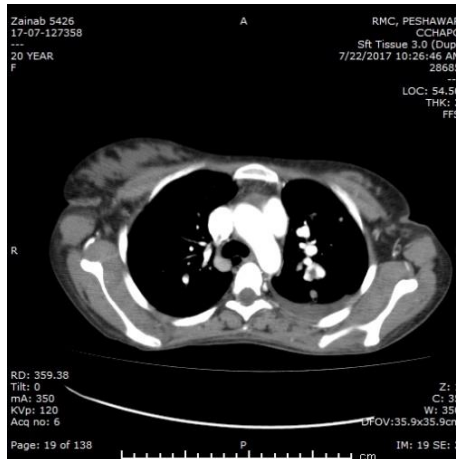
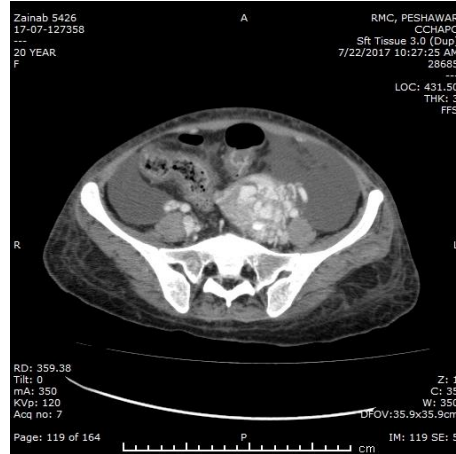
CASE REPORT

On 20 July 2017 a 20-year-old woman (Para 1) was referred from Hayatabad medical complex to Rehman medical institute. On the basis of her symptoms CBD stenting was tried in Hayatabad Medical Complex. She presented to RMI with jaundice, vomiting and diarrhea. The patient had a history of abortion and complicated pregnancy 2 years back, details of which are not available, and her surgical history was unremarkable. She was admitted and her further work up was done to find the cause of obstructive pattern of jaundice as suspected and anemia. She was

initially given supportive treatment with fluids, painkillers, PPIs and antibiotics to keep the patient stabilize and do further workup for the actual diagnosis.

On physical examination the patient was pale and in a general poor condition. Her abdomen was tender in right hypochondrium. Laboratory work up showed Hb of 9.5g/dl, wbc of $18.00 \times 10^9/l$, platelets of $115 \times 10^9/l$, HCG level of 2853.34 mIU/ml. Liver enzymes and total bilirubin was elevated (ALT 132U/l, ALP 505 U/l and bilirubin 2.31mg/dL) while serum albumin was low (1.9g/dl). Her kidney profile was normal (creatinine 0.53mg/dl and GFR 147.07ml/min/1.73sqm) and so was her coagulation profile with INR of 0.93.

Computed tomography (CT) scan of chest, abdomen and pelvis with contrast was done which revealed multiple small pulmonary nodules and aneurysms of peripheral small arteries in both lungs with no zonal predominance. Few of the aneurysms in right lower lobe and left lower lobe showed peripheral thrombosis. There were also small pleural effusions which was (L>R). There were no enlarged mediastinal lymph nodes. Abdomen showed a large aneurysm in pancreaticoduodenal region with peripheral thrombosis containing contrast pools. According to the report it appeared to be a large pseudoaneurysm with possible intrabiliary rupture causing obstructive jaundice. It had a peripheral hematoma containing contrast pools. It was also causing mass effect on duodenum and ampulla with resultant dilatation of biliary ducts and MPD. The aneurysm had feeders from branches of pancreaticoduodenal artery and celiac trunk. The CT also revealed a large mass in uterus associated with cystic component. It measured 7.9 x 5.7 cm.



On the basis of her history, examination and investigation the patient was diagnosed to have choriocarcinoma with metastasis to lungs and liver. She was admitted under interventional radiology unit for embolization of metastases of choriocarcinoma that has led to formation of arterioportal fistula with portal aneurysm. Her

embolization was planned. In the procedure the inferior pancreaticoduodenal feeders to the arterioportal fistula with aneurysm were embolized with 6mm x 14 mm coils. The superior pancreaticoduodenal and gastroduodenal artery was embolized with 6mm x 14 mm and 4mm x 10 mm (x2) .035 coils. Majority of the flow to the aneurysmal dilatation was stopped.

The patient was shifted and her vitals were monitored and post-procedure care was done. The next day the patient was examined. Her overall condition was improved and stable. A repeat CT scan was done to see the post procedure status of the aneurysm. The repeat CT revealed aneurysmal coiling with residual filling of the aneurysmal sac. Call to oncologist was sent for further management of the patient with chemotherapy.

DISCUSSION

GTDs are a heterogeneous group of malignant disorders that arise from the trophoblastic epithelium of the placenta. The characteristic of it is a distinct tumor marker (β -HCG)^{1,7,8,11,12,17,20,22,23,25}. GTDs are classified into at least five distinct groups on the basis of histopathology, cytogenetic, and clinical features¹². They are complete and partial hydatidiform mole, invasive mole, choriocarcinoma, placental site trophoblastic tumor and miscellaneous trophoblastic lesions. Out of these Choriocarcinoma is a rare, highly malignant neoplasm of a syncytiotrophoblastic origin among the GTDs. This tumor is known for its association with molar pregnancy, a rapid hematogenous spread to multiple organs and sites, high HCG levels and a good response to chemotherapy^{1,12,17,19,22}. It is preceded by several conditions as follows: 50% arise in molar

pregnancies, 25% arise after previous abortions, 23% arise in normal pregnancies and 3% arise subsequent to ectopic pregnancies¹¹.

Choriocarcinoma tends to metastasize early by hematogenous route. Widespread metastases to different sites and a high beta-HCG are characteristic of choriocarcinoma. The favored sites of involvement are the lungs (94% of all metastatic choriocarcinoma), vagina (44%), liver (28%) and brain (28%), followed by the skin, gastrointestinal tract, kidney, breast and bones^{11,24}. The clinical signs can be very different and vague depending on the site of the lesions. The disease most often presents with symptoms related to metastatic spread to different sites as the primary tumor may remain very small^{11,22}. Approximately out of all the patients with choriocarcinoma 30% of the patients show metastases at the time of diagnosis²⁰ and present with symptoms related to the site involved. In the case we described the patient presented with signs and symptoms of obstructive jaundice. On basis of her condition CBD stenting was done earlier to relieve her obstructive pattern but it did not show any significant results. After being referred her thorough history, examination and workup was done. On history the patient previously had a history of abortion and complicated pregnancy 2 years back. CT scan of chest, Abdomen and pelvis showed choriocarcinoma with metastasis to the lungs and liver mainly. The aneurysms in the lungs were small while findings in the liver were significant with formation of arterioportal fistula with portal aneurysm. Embolization of the aneurysm with coils was done and the patient was assessed. Call to oncologist was done for further management of the patient with chemotherapy.

	0	1	2	4
Age	<40	≥40	–	–
Previous pregnancy	Mole	Abortion	Pregnancy on term	–
Delay between the end of pregnancy and the beginning of chemotherapy	<4 mois	4–7	7–13	>13
Plasmatic HCG before the treatment (IU/l)	<10 ³	10 ³ –10 ⁴	10 ⁴ –10 ⁵	>10 ⁵
Larger tumor	–	3–5	>5	–
Metastatic sites	Lang (standard X Ray)	Kidney, spleen	Bowel, ileon	Brain, liver
Number of metastatis	0	1-4	5-8	>8
Failure of prior chemo therapy	–	–	Mono-chemotherapy	Poly-chemotherapy

Our patient was classified stage IV according to FIGO (distant metastases other than in lungs) and scored 11 points in the risk assessment scale. So she was urgently considered for chemotherapy.

CONCLUSION

The presentation of primary choriocarcinoma can be present. It may also be absent or subtle, possibly due to the spontaneous regression of the tumor itself, and the a metastatic lesion may presents as either a hypervascular mass at imaging studies, or progressive anemia, jaundice

or intra-abdominal hemorrhage, hemothorax etc depending upon the site involved.

Choriocarcinoma can be effectively treated with chemotherapy but in case where the main concern is incontrollable hemorrhagic complication and is the most common cause of death in patients, embolization seems to be an optimal and effective way of controlling and ceasing bleeding as compared to surgical method. After stabilizing the patient chemotherapy should be started to control the metastases and prevent its further spread.

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