Case Report

Choriocarcinoma syndrome in a case of abdominal testicular germ cell tumor with long-standing bilateral gynecomastia

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Abstract

Choriocarcinoma syndrome is an extremely rare, life-threatening condition seen in patients having advanced choriocarcinomas with large tumor burden. We report an unusual case of choriocarcinoma syndrome wherein a long-standing bilateral gynecomastia was ignored and an underlying testicular malignancy undiagnosed.

Keywords: Choriocarcinoma syndrome, computed tomography, gynecomastia, mammography

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Testicular choriocarcinomas are rare nonseminomatous germ cell tumors (NSGCTs) arising from gonadal germ cells, seen in men aged 20–40 years. Cryptorchidism, especially abdominal undescended testis, is associated with a greater risk of testicular malignancy. The incidence of pure choriocarcinomas is <1% of all testicular malignancies, and uncommon extragenital locations include retroperitoneum, lung, mediastinum, brain, and gastrointestinal tract.[1] About 60%–70% of the patients with testicular choriocarcinomas have advanced disease at presentation with symptoms of pulmonary and brain metastases including acute bleeding.[1,2]

Gynecomastia, the generalized enlargement of male breasts, though most commonly benign, can rarely occur as a manifestation of underlying testicular germ cell tumor (GCT).[1,2] We report an unusual case of abdominal testicular choriocarcinoma with long-standing benign gynecomastia suddenly developing choriocarcinoma syndrome. Literature search did not reveal any such prior case.

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A 41-year-old male with a history of breast enlargement for the last 3 years presented with cough and breathlessness of 7-day duration. Physical examination revealed a palpable mass per abdomen, empty right scrotal sac, and bilateral gynecomastia. Mammography revealed fibroglandular tissue radiating from the nipple into the deeper adipose tissue suggesting long-standing benign gynecomastia [Figure 1]. Computed tomography of the abdomen revealed a large necrotic soft-tissue attenuation mass in the abdomen, a single left-sided testis in the scrotum, bilateral gynecomastia, and multiple lung nodules [Figure 2]. Histopathology of the abdominal mass showed extensive necrosis and hemorrhage with cytotrophoblasts and syncytiotrophoblasts exhibiting abundant eosinophilic cytoplasm and hyperchromatic nuclei, the characteristic features of choriocarcinoma. Fine-needle aspiration cytology (FNAC) of lung nodules confirmed the lesions to be pure choriocarcinoma, and the FNAC from the breast was found to be benign hyperplasia. Blood biochemical workup revealed elevated human chorionic gonadotropin (HCG) levels (71,300 IU/L),

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further confirming the diagnosis. Chemotherapy with interval surgical debulking of the abdominal mass was planned, but the patient developed acute respiratory distress and succumbed within 2 weeks.

Choriocarcinoma is an aggressive malignancy characterized by several unique features distinct from other GCTs. These include rapid growth of tumor outgrowing the blood supply; tendency for angioinvasion; hematogenous spread of highly vascular metastases to lung, liver, brain, lymph nodes, and other organs; elevated serum HCG level (tumor marker and prognostic indicator); and sometimes, a small "burned out" primary lesion exhibiting histological regression in the presence of metastases.[1-3]

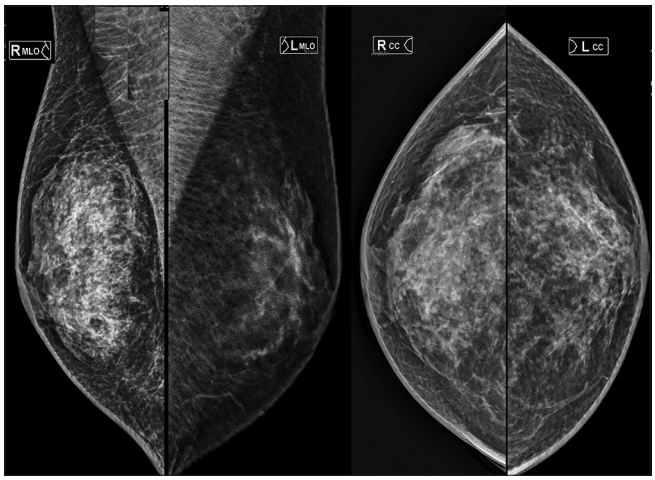

Figure 1: Mammographic images of both breasts in mediolateral oblique and craniocaudal views show enlargement of the breasts with diffuse density. No identifiable mass or secondary signs of malignancy are seen. The fibroglandular tissue seen radiating from the nipple and retroareolar area into the deeper adipose tissue is suggestive of long-standing gynecomastia

The International Germ Cell Cancer Collaborative Group classifies NSGCTs into good-risk, intermediate-risk, and poor-prognosis groups based on three criteria: the primary tumor site, serum tumor marker levels, and the presence of extrapulmonary metastases.[4] A subset of patients within the poor-risk NSGCTs (those who have pure choriocarcinoma, widespread metastases, and high HCG level) are prone to develop an extremely rare and fatal complication described as "choriocarcinoma syndrome."[4] Choriocarcinoma syndrome, originally reported by Logothetis in 1984, is a life-threatening condition seen in patients with advanced choriocarcinoma and large tumor burden. It can occur either spontaneously or shortly after administering chemotherapy, causing lethal hemorrhage from metastatic lesions and/or acute respiratory distress leading to rapid death.[3]

This is an extremely rare presentation of choriocarcinoma syndrome in a patient with long-standing gynecomastia that was ignored, and the intra-abdominal testicular tumor remained undiagnosed for years. This case underscores the fact that in patients with gynecomastia, underlying hormone-producing

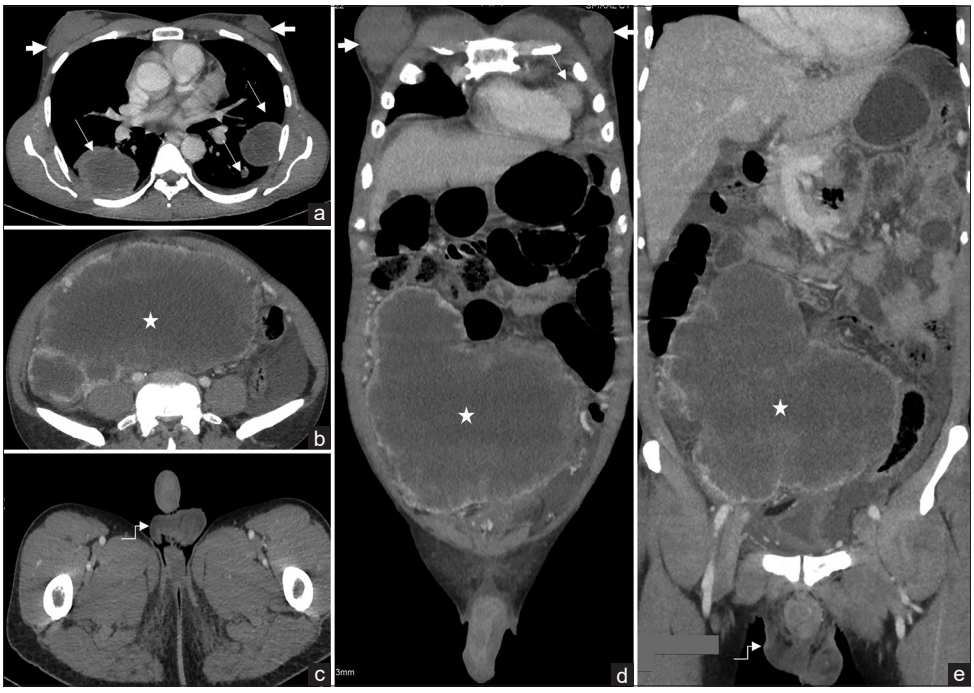

Figure 2: (a-e) Contrast-enhanced computed tomography axial and reformatted coronal images of the abdomen and chest reveal large lobulated peripherally enhancing soft-tissue attenuation mass with central necrosis (★) and multiple lung nodules (thin arrows) – biopsy-proven choriocarcinoma from an undescended testis with lung metastases. Bilateral gynecomastia (thick arrows) and empty right scrotal sac (elbow arrow) with a single left-sided testis are also seen

testicular malignancy should be considered and screened for avoiding delayed diagnosis and adverse outcomes.

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Conflicts of interest

There are no conflicts of interest.

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