

Cutaneous and systematic metastasis of testicular choriocarcinoma

Case report and literature review

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Abstract

Rationale: Cutaneous metastasis of testicular choriocarcinoma is exceptionally uncommon. To our knowledge, only 14 cases have been reported in the past 10 years in the pubmed. We have an uncommon case of testicular choriocarcinoma who has metastasized to the adjacent skin and organ systems.

Patient concerns: An 18-year-old male was diagnosed with initial presentation of cutaneous mass at the left back. Followly, biopsy was performed under local anesthesia. Histopathological examination was consistent with the diagnosis of metastatic choriocarcinoma.

Diagnoses: The histopathological assessment of the biopsied tissue, in combination with elevated serum β -HCG levels and presentation of testicular mass, indicated primary testicular choriocarcinoma with cutaneous and systemic metastasis.

Interventions: Subsequently radical orchiectomy was performed.

Outcomes: Despite the case completed one cycle of cisplatin-based regimen chemotherapy, he died of multiple organ dysfunction syndrome 2 months after surgery.

Lessons: In this report, cutaneous metastasis with testicular choriocarcinoma is extremely rare. It is important to recognize that this unusual variant of testicular choriocarcinoma has the potential to behave aggressively and to metastasize.

Abbreviations: β -HCG = beta-human chorionic gonadotropin, AFP = alpha-fetoprotein, CK = cytokeatin, CT = computed tomography, GTCs = germ cell tumors, LDH = lactate dehydrogenase, MODS = multiple organ dysfunction syndrome, MRI = magnetic resonance imaging.

Keywords: cutaneous metastasis, radical orchiectomy, rare case, systematic metastasis, testicular choriocarcinoma

1. Introduction

Choriocarcinoma is a clinically aggressive tumor with metastasis commonly occurring in the lungs and the brain. Cutaneous metastasis of choriocarcinoma is rare, and only a few cases have been reported in the literature to date.^[1] Germ cell tumors (GCT) account for 95% of testicular carcinoma. GCTs are divided into the 2 types of seminoma and non-seminoma, with the former mostly comprising malignant tumors arising in the testes and the

latter embryonal cell carcinoma, teratoma, yolk sac tumors, and choriocarcinoma.^[2,3]

Testicular choriocarcinoma is rare and accounts for <1% of testicular GCTs; it usually metastasizes to the lungs, liver, and brain.^[4] Testicular choriocarcinoma accompanied by cutaneous metastasis is especially uncommon.^[5,6] Here, we have presented a rare case of testicular choriocarcinoma accompanied by cutaneous and systematic metastases.

2. Case report

The relatives of the patient provided consent for the publication of this case report and for conducting any related studies.

On November 10, 2015, an 18-year-old boy was admitted to our institution with the chief complaint of bilateral chest pain lasting over 1 month. Physical examination revealed a dark-pigmented, soft, nodular swelling on the left back, measuring 2 cm \times 2 cm (Fig. 1), as well as a left testicular, non-tender mass that had been present for the last 3 months. A biopsy of the lesions on the left back was performed under local anesthesia. Histopathological examination was consistent with a diagnosis of testicular metastatic choriocarcinoma (Fig. 2).

The patient's serum beta-human chorionic gonadotropin (β -HCG) level was 10,000 IU/mL (normal, <5 IU/mL). Additional serum tumor markers, including alpha-fetoprotein (AFP) and lactate dehydrogenase, were within the normal ranges. Scrotal ultrasonography revealed that the non-tender mass in the left testis was a heterogeneous mass measuring 5 cm \times 5 cm \times 6 cm.

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Figure 1. The gross and microscopic appearance of metastatic lesion of testicular choriocarcinoma. There was a 2 × 2 cm dark-pigmented, soft, nodular swelling in the left back region (red arrow).

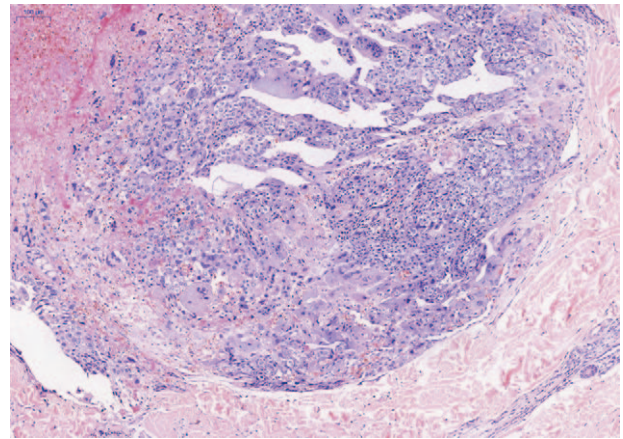


Figure 2. Histological examination showed that tumor cells were made up of syncytiotrophoblast and cytotrophoblast cells (H&E, × 100).

Contrast-enhanced abdominal and chest computed tomography (CT) scans revealed the presence of multiple bilateral pulmonary tumors (Fig. 3A) and extensive retroperitoneal metastatic lesions (Fig. 3B).

The histopathological assessment of the biopsied tissue in combination with the elevated serum β -HCG levels and the presentation of the testicular mass indicated primary testicular choriocarcinoma with cutaneous and systemic metastases. The patient underwent left radical orchiectomy under general anesthesia 5 days after the biopsy in our department. Gross pathological examination of the specimen revealed a nearly round and pinkish fibrous lesion with a dimension of 5 cm × 4 cm × 5 cm in the left testicular region (Fig. 4). Tumor cells comprised syncytiotrophoblast and cytotrophoblast cells; extensive hemorrhage, and necrosis were also observed. The results of immunohistochemical staining revealed that the tumor cells were strongly and diffusely positive for HCG and cytokeratin antigens and negative for AFP and vimentin. The final histopathological diagnosis was consistent with pure testicular choriocarcinoma (Fig. 5). The final pathologic stage was T₁N₂M_{1b}.

The patient's serum β -HCG level continued to remain at 10,000 IU/mL 1 week after the surgery. One cycle of chemotherapy with cisplatin, bleomycin, and etoposide was performed 2 weeks after the surgery; however, the condition of the patient deteriorated rapidly before a second chemotherapy cycle could be

administered. Meanwhile, he also presented with dizziness, vomiting, headache, and other symptoms consistent with central nervous system involvement. Simultaneously, the serum β -HCG level rapidly elevated to 15,000 IU/mL. A magnetic resonance imaging (MRI) scan revealed multiple cerebral metastatic tumors (Fig. 6). Further therapy was not performed at the request of the patient's relatives. The patient died of multiple organ dysfunction syndrome 2 months after surgery.

3. Discussion

The skin is an uncommon site for metastatic tumors, and the overall incidence of cutaneous metastasis of different tumor types is 0.7% to 9%. Common primary cancers causing cutaneous metastases are breast and colon cancer in women as well as lung and colon cancer in men; in both men and women. The most common sites of skin metastasis are the chest and abdomen, followed by the head and neck. Cutaneous metastasis of genitourinary tumors have been associated with cancers of the prostate,^[7] bladder,^[8] and kidney.^[9]

Choriocarcinoma has a marked tendency to metastasize early. Testicular choriocarcinoma that metastasizes to the skin has been

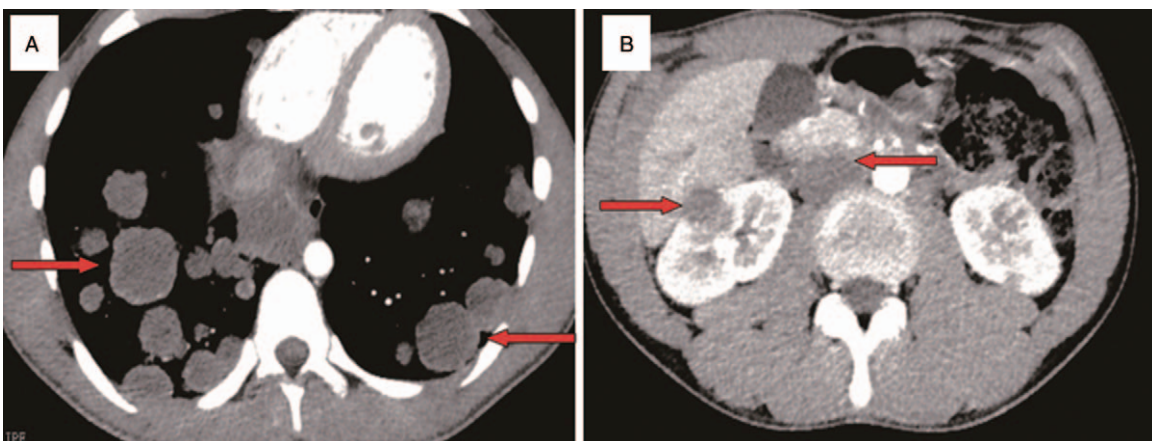


Figure 3. A, Contrast-enhanced CT scan demonstrated that there were bilateral pulmonary multiple tumor (red arrow). B, Contrast-enhanced CT scan demonstrated that there were extensive retroperitoneal metastatic lesions (red arrow).



Figure 4. The gross and microscopic appearance of testicular choriocarcinoma. Macroscopic examination revealed a roundish, pinkish, fibrous lesion (red arrow), measuring 5 × 4 × 5 cm in dimension.

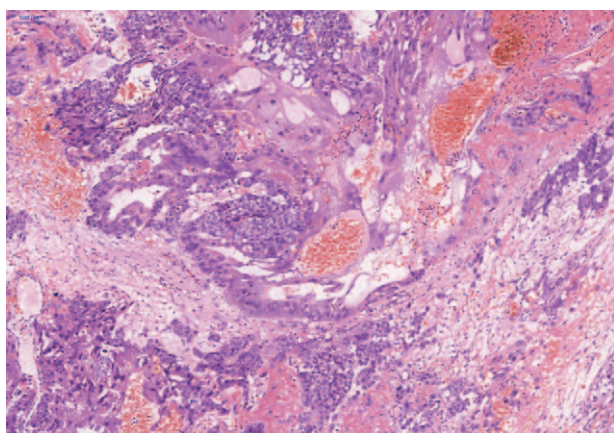


Figure 5. Histological examination were compatible with a diagnosis of testicular choriocarcinoma (H&E, × 100).

rarely reported, and, to the best of our knowledge, only 14 cases of metastatic choriocarcinoma with skin metastasis have been reported in the English literature to date.^[10,11] The present case is very interesting not only because the incidence of such tumors is extremely low but also because this is the first case of testicular choriocarcinoma accompanied by cutaneous metastasis reported in China.

Choriocarcinoma is a malignant growth of trophoblastic cells, which is characterized by the secretion of β -HCG. It usually develops as gestational choriocarcinoma, originating from fetal trophoblasts associated with a previous hydatidiform mole pregnancy.^[12] Rarely, it originates from germ cells in the testes or ovary. Choriocarcinoma is conventionally classified as pure choriocarcinoma, which is composed of only syncytiotrophoblastic and cytotrophoblastic components, and as mixed GCT, including choriocarcinoma as one component. The present case is markedly different from previously reported cases because the metastasis was widespread and consisted of retroperitoneal and bilateral pulmonary lesions; moreover, multiple cerebral lesions caused by pure testicular choriocarcinoma were seen. The diagnosis of metastatic choriocarcinoma was established according to the patient's medical history and radiological results; it was further confirmed by the histological analysis.

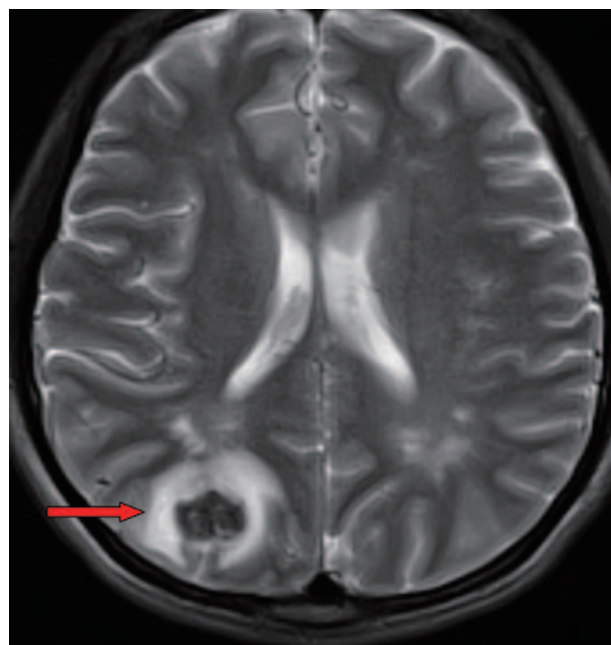


Figure 6. MRI demonstrated that there were multiple cerebral metastatic tumors (red arrow).

This case report highlights that suspicious skin lesions should be investigated thoroughly by a dermatologist to assess any underlying malignancies. Furthermore, serum β -HCG levels may provide important clues regarding the response to therapy and tumor progression. Similarly, in the present case, β -HCG levels increased after 1 chemotherapy cycle. This coincided with clinical progression evidenced by further examinations revealing multiple cerebral lesions. Thus, in such a case, it may be prudent to monitor the symptoms corresponding to the functioning of the central nervous system and promptly perform brain imaging.

Testicular choriocarcinoma with cutaneous metastasis has a poor prognosis because it is associated with an advanced stage of the disease. Although the present case was actively treated, including the resection of the primary lesion and adjuvant chemotherapy, the patient unfortunately died of the disease 2 months after surgery due to the highly aggressive and malignant nature of the disease. The results of this case are consistent with those of similar previously published cases; however, an increase in the number of case reports will help improve the diagnostic criteria, prognostic factors, surveillance, and therapeutic modalities.

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