A Case of Choriocarcinoma with Lung Metastasis as the First Symptom

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Case Report

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Abstract

Background: Pure testicular choriocarcinoma is very rare in clinic and the patient's age is 20-39 years (median age 29 years). Because of its polymorphic manifestations, it is not always suspected and patients are sometimes misdiagnosed. The case should be reported due to the rarity of the disease, the onset age and the rapid progression of the disease. This case with primary lung cancer as the first diagnosis is a misdiagnosis. We hope we can provide some experience for clinical diagnosis and treatment.

Case presentation: We report the case of a 46-year-old man who was admitted to our hospital with bloodshot sputum. Histopathological and immunohistochemical examination revealed pure choriocarcinoma. The patient had received radiotherapy, chemotherapy, immunotherapy. Over the next 5 months, the patient developed symptoms of multiple system metastases and died soon.

Conclusions: Testicular choriocarcinoma is progressing rapidly. The clinical feature of choriocarcinoma is hemorrhaging at the metastatic site. Early diagnosis and timely treatment should be combined with the clinical manifestations and medical history of patients to strive for a longer survival time. We hope this case can provide guidance for the clinical diagnosis and treatment of the disease.

Background

Pure testicular choriocarcinoma is very rare in clinic and the patient's age is 20–39 years (median age 29 years). Because of its polymorphic manifestations, it is not always suspected and patients are sometimes misdiagnosed. Distant metastasis may be characteristic of choriocarcinoma. The clinical feature of choriocarcinoma is hemorrhaging at the metastatic site, which is an acute medical emergency with a high morbidity and mortality. Treatment should aim at the goal of normalization of tumor markers, and in refractory cases, advanced ventilator and innovative techniques should be taken into account, preferably in the setting of clinical trials. Due to the rarity of the disease, the onset age and the rapid progression of the disease, we decide to report this case. This case with primary lung cancer as the first diagnosis is a misdiagnosis. We hope we can provide some experience for clinical diagnosis and treatment.

Case Presentation

A 46-year-old male who was found with bilateral lung metastases and intrapulmonary infection during chest Computed Tomography (CT) examination at the "Second People's Hospital of jimo city" on October 17, 2018 due to blood streaks in sputum, had no chest pain, chest distress, obvious yellow sputum and history of fever and night sweats, and was not treated. Three days later, a whole chest and abdomen enhanced CT was performed in the thoracic surgery department of Qingdao Municipal Hospital, which showed that malignant stromal tumors of the small intestine with multiple metastases to both lungs, liver and right kidney were considered. (As shown in Fig. 1). Video-assisted thoracoscopic pulmonary wedge

resection on 26 October 2018. Paraffin pathology revealed an infiltration of choriocarcinoma tissue in the right lower lobe lung, and immunohistochemical results: β -HCG(+), CK5/6(-), P63(Partial+), P40 (Partial+), Ki67(80%+), TTF-1(-), NaPSINA (-), Pax-8(-), CEA(-), CK7 (+), NSE (-), CK(+)(Fig. 2). Based on our patient's pathological findings, immunohistochemical and congenital cryptorchidism history, and our patient's clinical presentation, we present the diagnosis of pulmonary metastasis from pure testicular choriocarcinoma. The patient was transferred to the First Oncology Department of Qingdao Municipal Hospital for internal medicine comprehensive treatment on November 05, 2018. He suffered from moderate anemia and was given blood transfusion treatment. After the general condition was improved, chemotherapy was started on November 13, 2018. The chemotherapy plan and dose were changed several times due to the progression of the patient's condition. The patient received several chemotherapy plans successively, including vincristine + Tegafur Gimeracil Oteracil Potassium Capsule(S-1), vincristine + S-1 + dactinomycin, vincristine + etoposide + dactinomycin + S-1, and methotrexate + cisplatin, with unsatisfactory results. His general condition was declining with persistent melena, hemoptysis, and anemia. An imaging examination was performed on January 16, 2019, which revealed brain metastasis (Fig. 3), and whole brain radiotherapy was started: 4000 cGy/20 f. Xindilizumab 3-cycle immunotherapy was carried out from 2019-03-01 to 2019-04-22. From 2019-05-19, the patient's condition progressed, melena persisted, and anemia did not improve. In the end stage of the disease, the patient was treated with transfusion of leukocyte-removed suspended red blood cells. However, his general condition was still unable to be improved, and his general condition was poor. His family members abandoned further treatment, and he was discharged automatically and died soon after.

Discussion And Conclusions

Testicular germ cell tumors are a collection of tumors that originated from the primordial germ cells in the gonads of embryos and form during their differentiation, maturation, and migration. Choriocarcinoma is the fruit of the extraembryonic differentiation of primitive pluripotent undifferentiated germ cells. Pure choriocarcinoma of the testis is very rare clinically, accounting for less than 1% (0.19%) of testicular germ cell tumors, but it is mixed with other germ cell tumor components in 8% of testicular germ cell tumors. The incidence of testicular choriocarcinoma is 2–4/10 million, accounting for 0.01%-0.02% of male malignant tumor¹. Male choriocarcinoma usually occurs at the age of 20–39 years². The cause of testicular choriocarcinoma is still not clear, and there are many risk factors according to epidemiological analysis. It has been reported in the literature that the chance of tumor occurrence in patients with cryptorchidism is 20–40 times higher than that in patients with normal testis³. The reason is not only in the testis itself, but also related to family genetic factors, gonadal agenesis, endocrine disorders, etc. Genetic studies have shown that testicular tumors are associated with short-arm heterotopic chromosome 12, and changes in p53 gene are also associated with the development of testicular tumors⁴. The choriocarcinoma of the testis is the most aggressive and fastest-growing germ cell tumor. At the time of diagnosis, most cases had been transferred. Because of the high degree of vascularization of these lesions, patients usually present with symptoms of bleeding at the metastatic site⁵. Choriocarcinoma is usually characterized by early hematogenous spread to the lung and brain⁶.

Gastrointestinal metastases are extremely rare. Most metastatic lesions of the gastrointestinal tract are found in the small intestine⁵. The main symptoms of gastrointestinal metastases are melena and/or hematemesis and anemia. Because the most common sites of metastasis are the lungs and brain, patients usually present with seizures, stroke-like symptoms, confusion, and/or hemoptysis. Imaging lacked characteristic changes that could be distinguished from other types of germ cell tumors. We report the case of a 46-year-old man with multiple distant metastases, an age at which choriocarcinoma is rarely seen. Due to the rarity of the case, information on the treatment and follow-up of choriocarcinoma is limited. Choriocarcinoma is less sensitive to chemotherapy. Most cases progressed so rapidly in clinical presentation that the patient did not respond to standard chemotherapy, which included 3 or 4 cycles of BEP (bleomycin, etoposide, and cisplatin)⁷. The treatment of nonpregnant choriocarcinoma from the perspective of immunotherapy, cancer cells are from their own, poor immunity, the remaining cells after chemotherapy can not be controlled and easy to relapse, the mortality rate is high⁸. Due to early hematogenous dissemination, choriocarcinoma has a worse prognosis than other testicular tumors. Studies have suggested that the higher serum human chorionic gonadotrophin (HCG) may reflect the larger tumor size and worse prognosis. Of the 14 patients reviewed by Sahraoui et al.⁹, 4 died 3 d to 14 months after treatment and 11 survived 6 to 96 months. Requena et al.¹⁰ reported the 2-year tumor-free survival of a patient with pure choriocarcinoma of the testis with brain, lung, and skin metastasis treated with radiotherapy and multiple courses of chemotherapy. For the pure choriocarcinoma of testis discovered at an early stage, because the metastases in other parts of the body are very small, it is possible to obtain a long-term tumor-free survival or even cure by orchiectomy on the diseased side and combined chemotherapy mainly with cisplatin. As choriocarcinoma of the testis is mainly metastasized through blood, chemotherapy-based comprehensive treatment can only be adopted for choriocarcinoma of the testis with early metastasis. Literature reports are inconsistent as to whether metastases need to be resected after chemotherapy. Some people¹¹ believe that residual lesions must be resected as much as possible. However, some scholars¹² believe that it is not necessary to resect metastases. Because choriocarcinoma is prone to multiple metastases throughout the body, it is difficult to achieve tumor-free survival. The diagnosis of metastatic choriocarcinoma in this patient was based on her medical history and imaging findings; This was further confirmed by immunohistochemistry. The regret of this case lies in the fact that the blood HCG content was not detected during the treatment, and our patient's CT report showed a small intestinal stromal tumor. In view of our patient's clinical manifestations and the characteristics of the disease, we suspected that the tumor at the small intestinal site was a metastatic tumor. As our patient's condition progressed rapidly and he was generally in a poor condition, biopsy confirmation could not be obtained by puncture. Approximately seven months from discovery to death, our patient presented with bloody sputum, chronic blood loss in the digestive tract, severe anemia, and intermittent blood transfusion therapy, which are confirming the clinical features of the disease as hemorrhage at the metastatic site.

This case report highlights the rapid progression of choriocarcinoma of the testis, and our patient unfortunately died of this disease within 1 month after giving up treatment due to the highly aggressive and malignant nature of the disease. The results of this case were consistent with those of similar

previously published cases; However, an increase in the number of case reports will help improve diagnostic criteria, prognostic factors, monitoring and treatment modalities. Testicular choriocarcinoma is progressing rapidly. Early diagnosis and timely treatment should be combined with the clinical manifestations and medical history of patients to strive for a longer survival time. We hope this case can provide guidance for the clinical diagnosis and treatment of the disease.

Abbreviations

1. CT: Computed Tomography
2. S-1 Tegafur Gimeracil Oteracil Potassium Capsule
3. BEP bleomycin+ etoposide+ cisplatin
4. HCG human chorionic gonadotrophin

Declarations

ETHICS APPROVAL AND CONSENT TO PARTICIPATE:

This case report has been approved by the Medical Ethics Committee of Qingdao Municipal Hospital.

CONSENT FOR PUBLICATION:

We are very grateful to the patient's family for permission to publish this case report, and written informed consent for publication of their clinical details and clinical images was obtained from the relative of the patient. A copy of the consent form is available for review by the Editor of this journal.

AVAILABILITY OF DATA AND MATERIAL:

We confirm the authenticity and availability of patient data. The datasets during the current study are not publicly available due individual privacy could be compromised, but are available from the corresponding author on reasonable request.

COMPETING INTERESTS:

The authors declare that there are no financial or other relationships that might lead to a conflict of interest of the present article.

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"TJ collected case data,analyzed and interpreted the patient data regarding the choriocarcinoma of the testis and was a major contributor in writing the manuscript. XYN,WJ and JCL performed the histological

examination of the choriocarcinoma, ZTL analyzed the imaging data of the patient, GCY is corresponding author. All authors read and approved the final manuscript."

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Figures

Fig.1

Figure 1

After the patient was admitted, the auxiliary examination was improved, A whole chest and abdomen enhanced CT showed that malignant stromal tumors of the small intestine with multiple metastases to both lungs, liver and right kidney were considered. However, the primary lesion did not have any symptoms.

Fig.2

Figure 2

Pathology of paraffin wax revealed infiltration of choriocarcinoma in the right lower lobe lung, and recommended to examine the testis for exclusion of metastasis. Immunohistochemical results: β -HCG(+), CK5/6(-), P63(Partial+), P40 (Partial+), Ki67(80%+), TTF-1(-), NaPSINA (-), Pax-8(-), CEA(-), CK7 (+), NSE (-), CK(+).

Fig.3

Figure 3

Two months after treatment, the patient developed symptoms of dizziness, and brain CT indicated brain metastasis.