Choriocarcinoma Syndrome of Primary Pulmonary Choriocarcinoma after Lung Lobe Resection: A Case Report and Review of the Literature

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Abstract

Introduction: Primary pulmonary choriocarcinoma (PPC) is extremely rare, especially in males. It is characterized by a poor response to therapy and shortened survival times. Here, we report a PPC case which was complicated by choriocarcinoma syndrome (CS) and a review of the literature.

Case presentation: A 59-year-old man with 30-pack-year smoking history was referred to our hospital because of hemoptysis and chest pain. Positron emission tomography-computed tomography (PET-CT) showed multiple bilateral pulmonary nodules and multiple metastases throughout the body. The patient underwent single-port thoracoscopic wedge resection of the right lung upper lobe. Histological examinations verified the diagnosis of choriocarcinoma. Three days after the operation, the patient developed massive hemoptysis. A bronchoscopic examination showed bleeding from the bilateral main bronchus. Despite management in the intensive care unit (ICU), the patient died two weeks after surgery.

Conclusions: A rapid and correct diagnosis of PPC is necessary. When the patient has massive hemoptysis, we should be alert to the occurrence of CS. Surgery and modified chemotherapy, based on the physical condition of the patient, may currently be the best therapy for PPC.

1. Introduction

Choriocarcinoma is a highly malignant tumor which originates from anaplastic tropho-blastic tissue. Most of the choriocarcinoma usually occur in the female reproductive tract after a gestational event[1]. Extragonadal choriocarcinoma, which accounts for only 2% to 5% of all germ cell tumors[2], generally occurs in the midline of the body, such as the retroperitoneum, mediastinum, pineal gland, and intracranially. Primary pulmonary choriocarcinoma (PPC) in males is easy to misdiagnose or delay the diagnosis; therefore, a potentially curative chemotherapy or surgery may also be delayed[3]. Furthermore, choriocarcinoma syndrome (CS) is a lethal hemorrhagic comolication, containing high-volume choriocarcinomatous elements and markedly elevated β-HCG [1, 4]. Herein, we reported a 59 years old male with multiple bilateral pulmonary nodules who underwent a surgical wedge resection of the right upper lobe. The diagnosis of PPC was confirmed by its morphology and immunotype combined with the postoperative β-HCG level. Unfortunately, the patient developed choriocarcinoma syndrome postoperative and progressed to multiple organ failure rapidly. Despite blood transfusion and respiratory support in the intensive care unit, the patient died two weeks after surgery. Next, we have described the clinicopathological features, treatment, and prognosis of PPC and CS with a short review of the literature.

2. Case Presentation

A 59 years old man complaining of hemoptysis and chest pain was admitted to our hospital. Four months ago, Chest computed tomography (CT) revealed a 43×26 mm parenchymal nodule in the posterior right upper lung lobe accompanied by emphysema, while the patient did not pay attention. One month ago, the patient had hemoptysis without obvious inducement, accompanied by progressive

shortness of breath, drenching night sweats, diminished appetite and a 5-kg weight loss, chest pain and fever. The chest enhanced CT scan showed that the right lung lesion was significantly larger than previous, and there were multiple nodules in both lungs. The patient has a history of smoking for 30 years (20 cigarettes per day) and drinking for 40 years (200g per day).

After admission, genital exam did not reveal any scrotal mass. The remainder of the physical examination was unremarkable. Chest x-ray was significant for bilateral pulmonary nodules. Doppler ultrasound of the testes and mammography of breasts were negative for malignancy. To determine whether the tumor occurred primarily within the lung systemic screenings were performed by positron emission tomography-computed tomography (PET-CT). The results showed that multiple high metabolic nodules throughout the body (Fig 1), multiple bilateral pulmonary nodules (Fig 2), especially large mass in the upper lobe of the right lung (Fig 3), a 22 mm metastatic lesion in the pancreas (Fig 4), and multiple bone metastases (Fig 5). The patient underwent single-port thoracoscopic wedge resection of the right lung upper lobe. Extensive metastases in the right lung were observed during the operation, with tumor diameters ranging from 2-6cm. Histopathologic workup of excised tumour showed poorly differentiated carcinoma and hemorrhage, with the few viable islands demonstrating syncytiotrophoblastic and cytotrophoblastic like cells (Fig 6). Immunohistochemical phenotype of pathological sections was that CK7(+),TTF-1(-),CK(+),CD34(-),CD117(-),OCT3/4(-),SALL4(+),HCGα(+),Ki67(+90%),Vimentin(-). Thus, we performed the serum β-HCG levels examination and the result was >10000.00(mlU/ml). The final pathological diagnosis was choriocarcinoma that met the diagnostic criteria for PPC.

Three days after the operation, the patient suddenly appeared consciousness disorder, sweating profusely and pinpoint pupils. Heart rate of the patient increases and the blood oxygen saturation continues to decrease. The patient was admitted to the ICU with tracheal intubation for respiratory support. A large amount of bloody fluid was sucked out of the bilateral main bronchus using bronchoscopy. The chest radiograph showed that multiple cluster and nodular shadows scattered in both lungs. Laboratory data was significant for white blood cell 13.85 900/mm which prompt the pneumonia. Subsequently, the patient developed cardiac dysfunction, manifested by increased myocardial markers. Cardiac color doppler ultrasound revealed that ventricular wall motion is not coordinated; left ventricular diastolic function decreased; ejection fraction was 60%. Simultaneously, the patient suffered acute renal failure, the endogenous creatinine clearance rate continued to decrease, to a minimum of 10.01 (ml/min/1.73m^2), and urea and creatinine continued to rise. Despite supportive treatment, the patient's condition continued to deteriorate. Due to multiple organ failure, the patient has no indications for chemotherapy and eventually died. Diagnosis of death: 1. Pulmonary choriocarcinoma with metastasis to the lung, brain, bone, pancreas, and lymph nodes 2. Severe pneumonia and respiratory failure 3. Acute renal failure; 4. Acute coronary syndrome.

3. Discussion

Due to infrequency of PPC, diagnosis before surgery is difficult [5]. In our case, the patient's single parenchymal nodule rapidly progress to multiple bilateral pulmonary nodules combined with multiple

metastases throughout the body during three months. It was not until postoperative pathology and β-HCG examination that we finally established the diagnosis of PPC. Therefore, a rapid and correct diagnosis of choriocarcinoma is necessary. The diagnostic criteria for PPC includes the lack of a previous urological malignancy, solitary or predominantlung lesion without a primary gonadal site, elevated serum β-HCG levels that normalize following surgery or chemotherapy, and pathologic confirmation of the disease[6]. In our case, genital exam and doppler ultrasound failed to find the lesions of the reproductive system. Nevertheless, the possibility of a metastasis from occult lesions in other locations could not be ruled out completely because the request for an autopsy was denied.

CS occurs shortly after the administration of chemotherapy and is connected with a high risk of fatal bleeding from metastatic lesions and frequently with acute respiratory failure with a high mortality rate at an early phase of the treatment induction [7]. Mostly, the hemorrhage appears shortly after the introduction of the chemotherapy, but there are also cases with pretreatment onsets reported in literature[8]. The basic mechanism of choriocarcinoma syndrome is probably massive lung metastases, massive intra-alveolar tumor-lysis, early necrosis of tumor cells, and consecutive superinfection, which can lead to acute respiratory failure (ARDS)[9]. Releasing of the cytokines is probably raising to systemic inflammatory response directing to multiorgan failure[10]. However, the cause of this patient's CS may be the surgery instead of chemotherapy y. There also have been report of CS after surgery, but the CS in that case occurred one and a half months after surgery[1], while the CS in our case developed only three days after surgery. This prompts us to diagnose PPC through minimally invasive or even non-invasive means as much as possible to avoid inducing CS. Especially when we found that the patient has massive hemoptysis, we should be alert to the occurrence of CS. We can use chemotherapy including bleomycin, etoposide, cisplatin, surgery and postoperative radiotherapy, and the prognosis will be improved[11].

In conclusion, PCC is highly malignant, progresses rapidly, and often has blood metastases at the time of diagnosis, and the prognosis is poor. The comprehensive treatment method of surgery combined with chemotherapy is advocated, but the effect is not satisfactory. Through clinical combined imaging and pathological examination, early detection, early diagnosis and early treatment are the keys to improving the prognosis of this disease.

Declarations

Statement

Written informed consent was obtained from the participant and his family members before the study began. All the investigation got the approval by Medical Institutional Review Board of West China Hospital of Sichuan University. The authors declare that they have no competing interests. There is no funding for this investigation.

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Author Contribution

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Figures
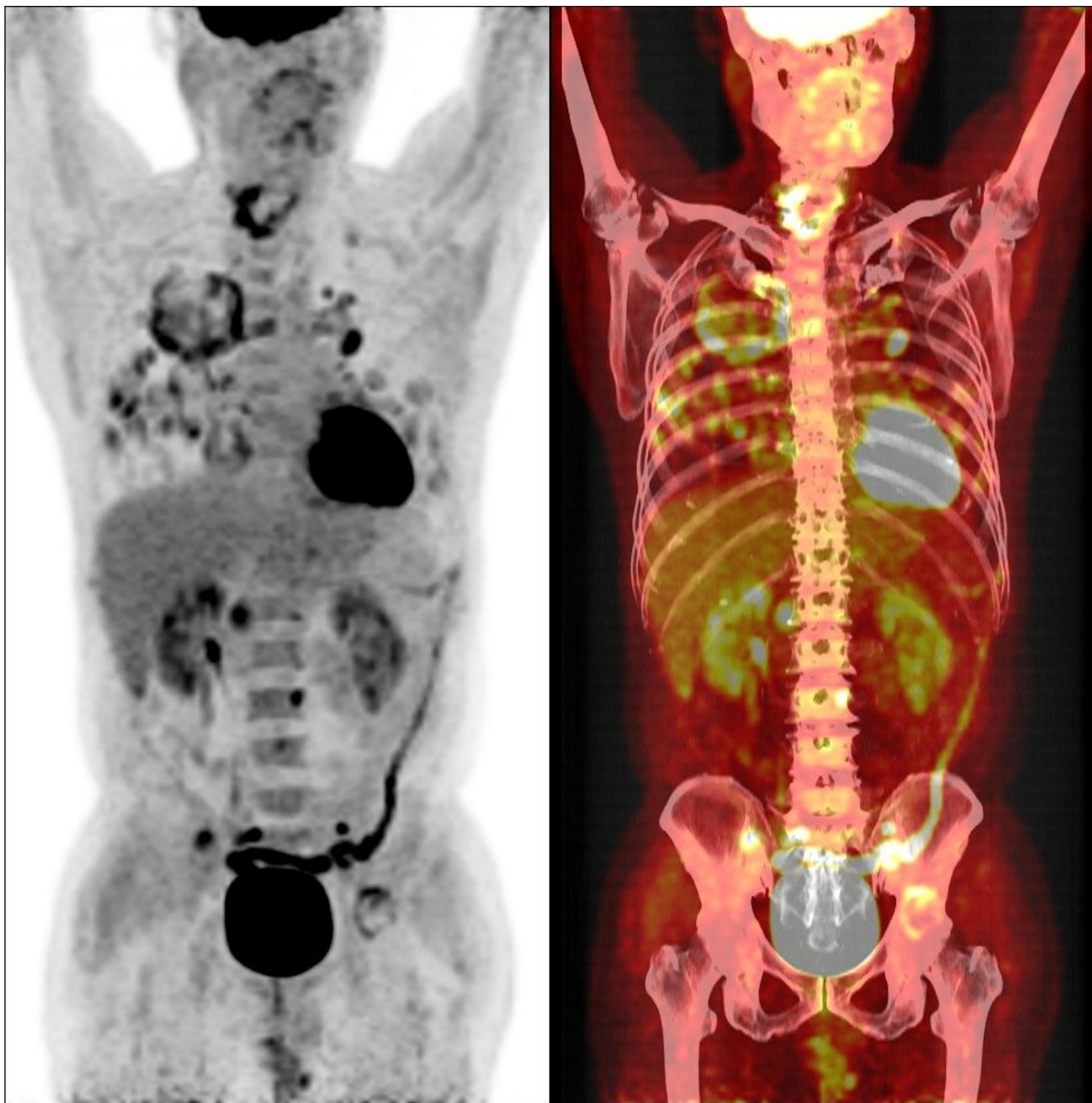
Figure 1

PET-CT revealed that multiple high metabolic nodules throughout the body.
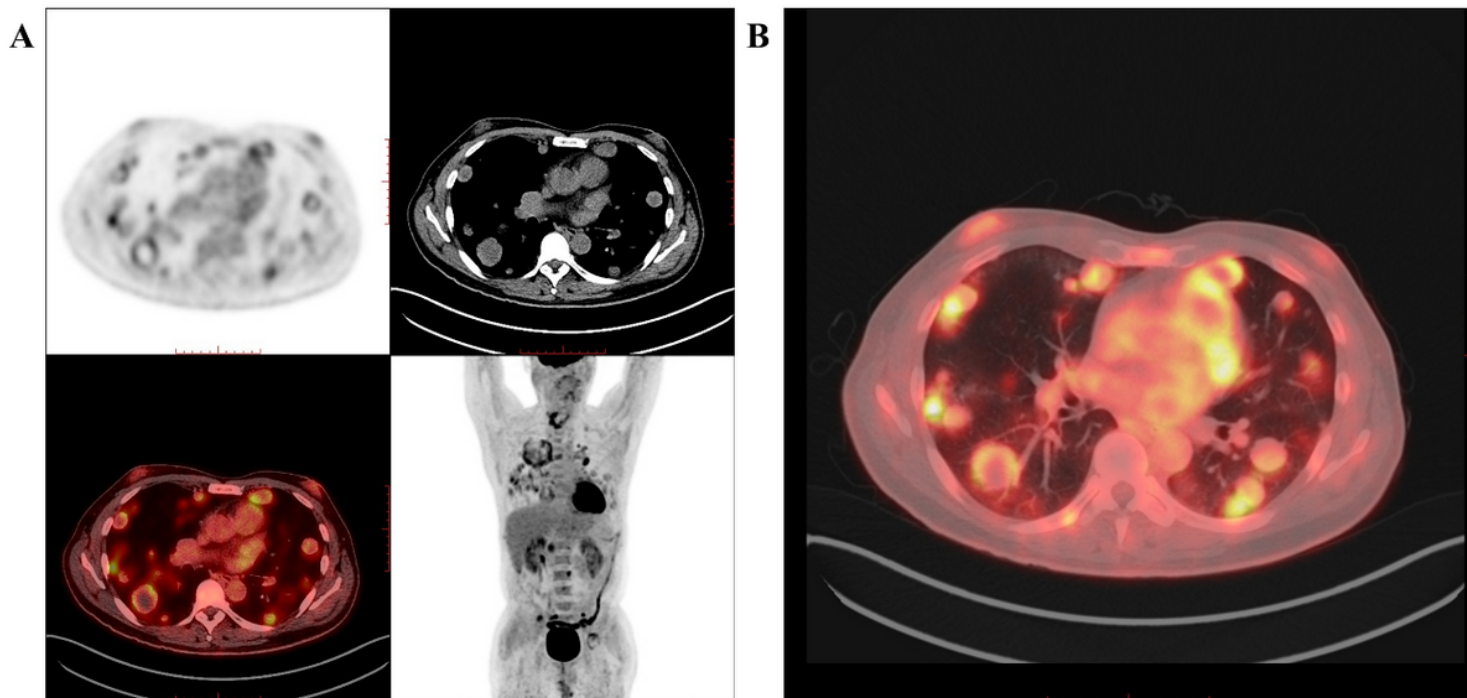
Figure 2

PET-CT revealed that multiple bilateral pulmonary nodules.
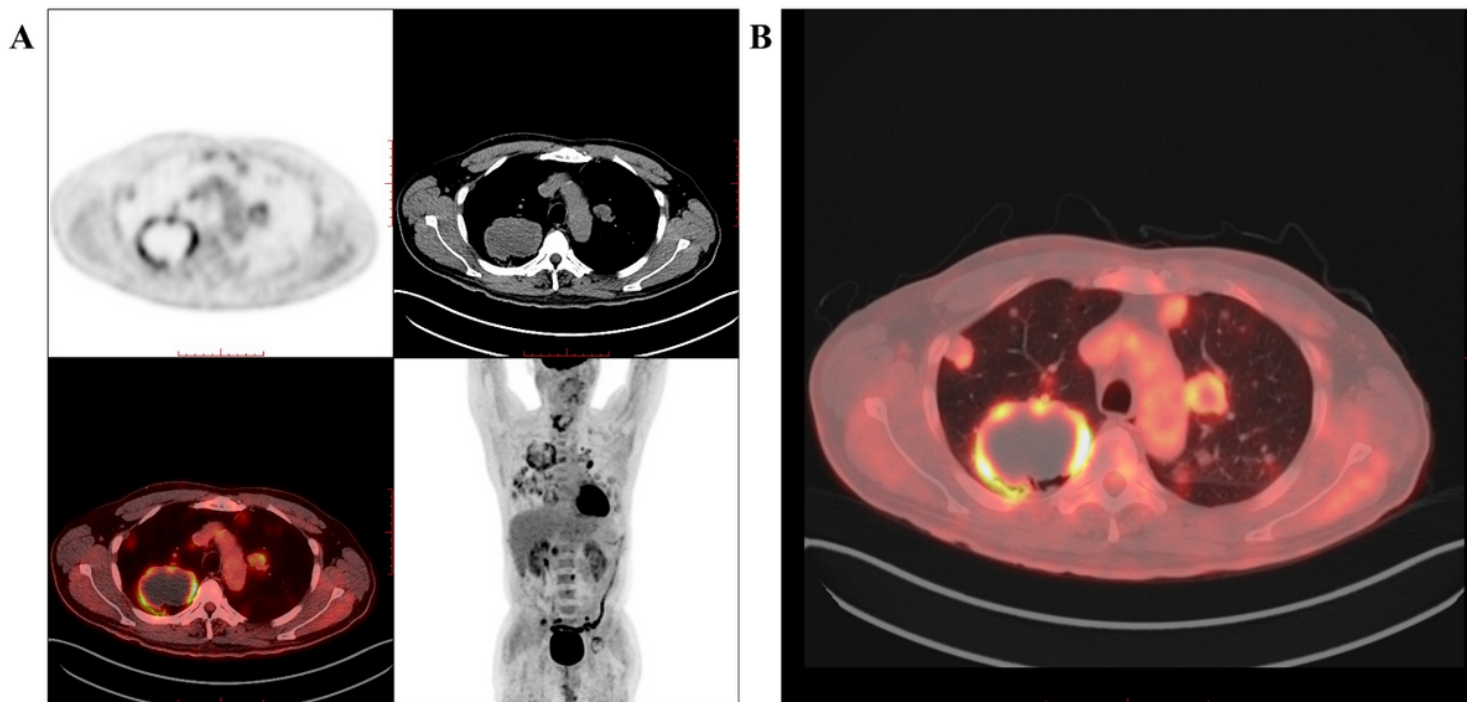
Figure 3

PET-CT revealed that large mass in the upper lobe of the right lung
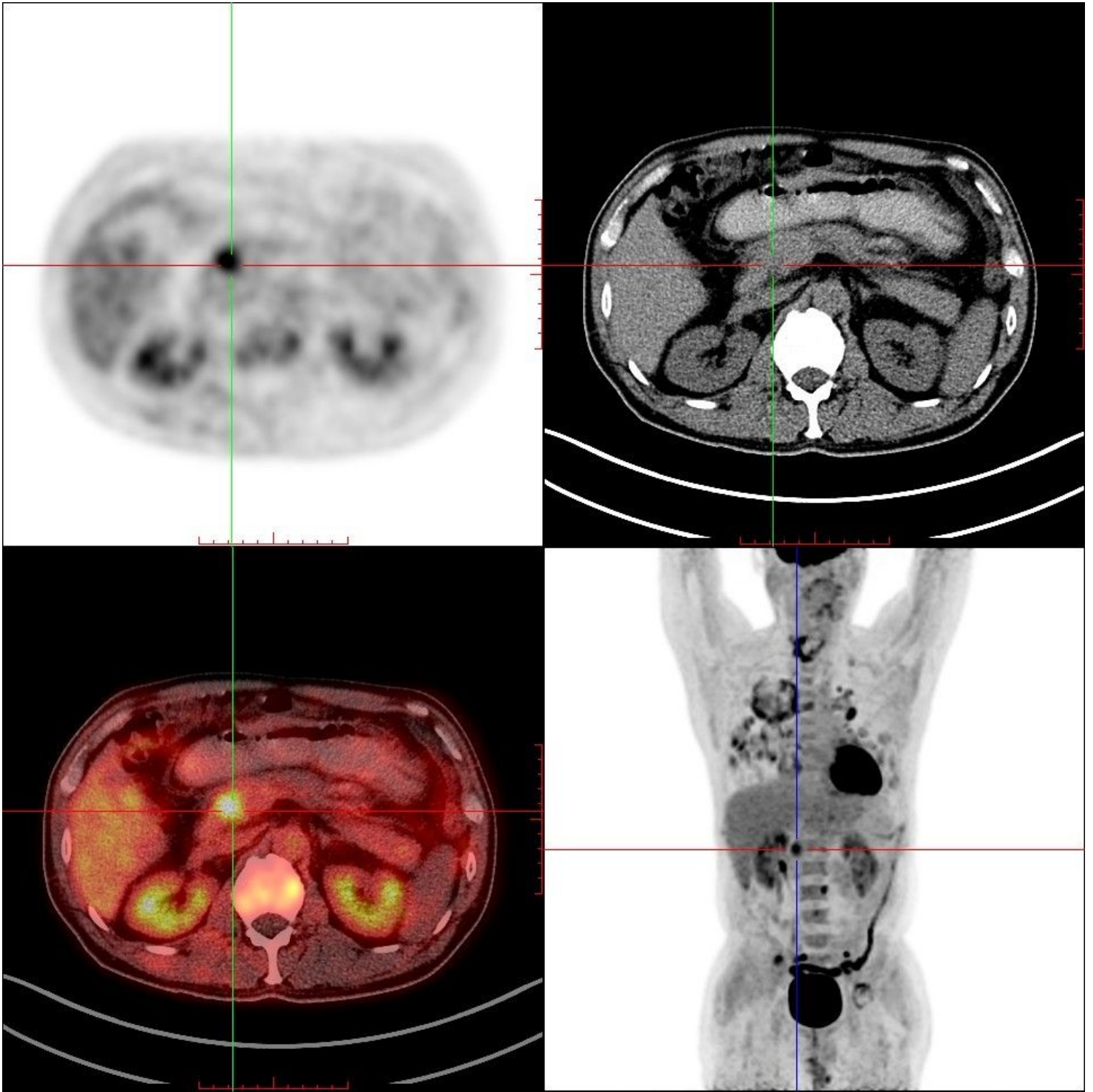
Figure 4

PET-CT revealed a 22 mm metastatic lesion in the pancreas
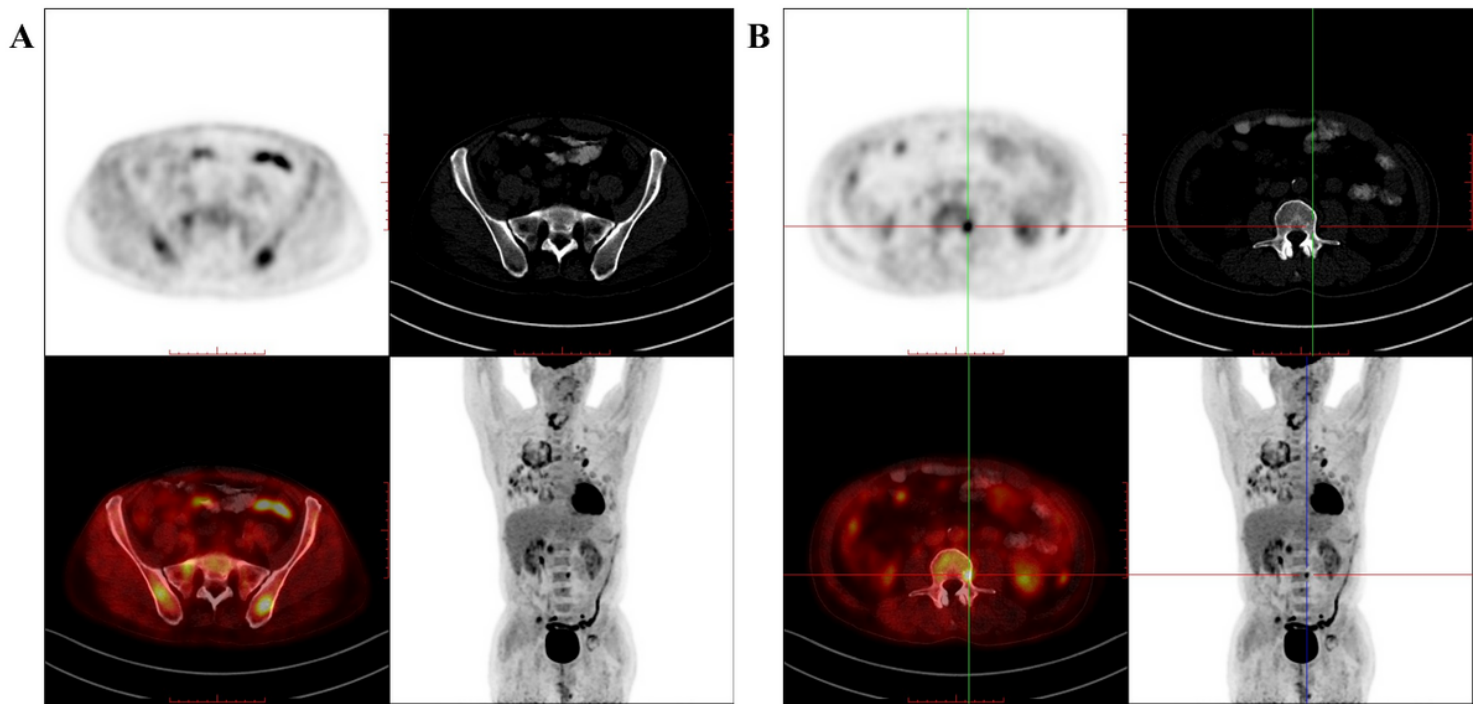
Figure 5

PET-CT revealed bone metastases in pelvis (A) and vertebra (B).
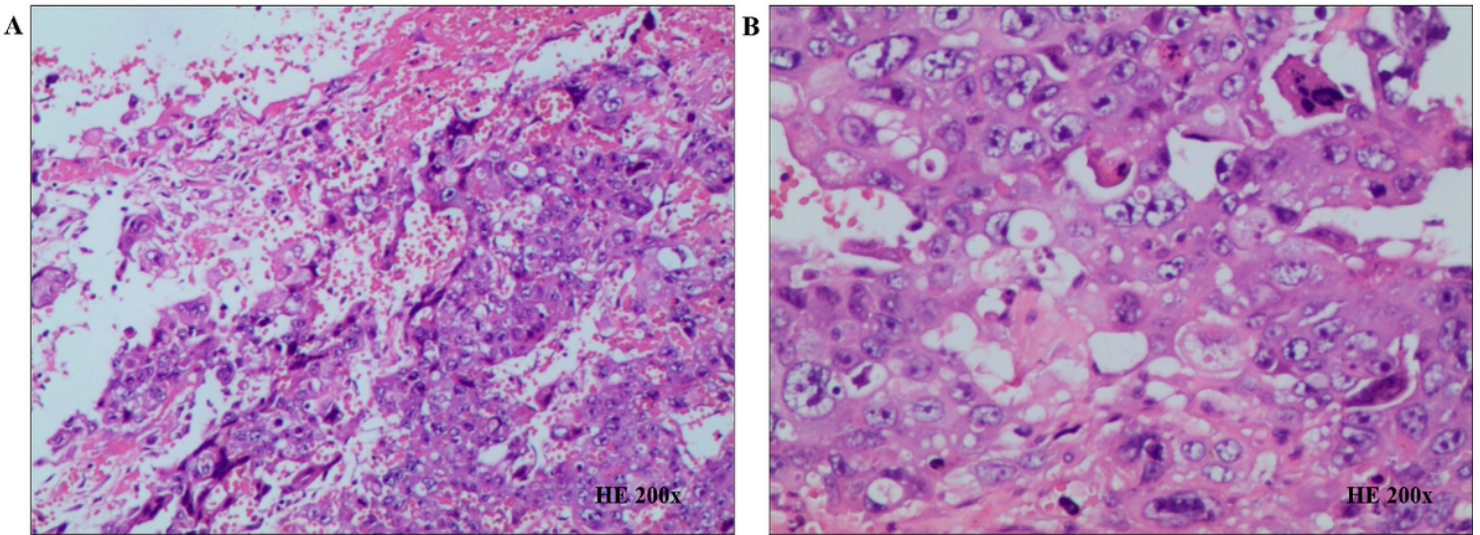
Figure 6

Histologic findings showing that the tumor was consisted of the poorly diffenrentiated adenocarcinoma, hemorrhage and islands demonstrating syncytiotrophoblastic and cytotrophoblastic like cells (hematoxylin and eosin stain; original magnification, ×200 and ×400).