

# Lumbar Vertebrae and Epidural Space Metastases of Choriocarcinoma: A Case Report

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## Case report

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# Abstract

**Background:** Choriocarcinoma is a rare and highly malignant tumor. It is characterized by the production of large amounts of human chorionic gonadotropin (HCG). The lumbar vertebrae and epidural space metastases of choriocarcinoma are extremely rare. This is a rare case report of a middle-aged woman with metastatic choriocarcinoma in the lumbar vertebrae, epidural space.

**Case presentation:** The present case report describes a 42-year-old Chinese female patient who was admitted to the spine surgery department of The Second Affiliated Hospital of Dalian Medical University with signs of low back pain for ten days, weakness in both lower limbs and dysuria for four days in 2019. Magnetic resonance imaging revealed an inhomogeneous nodular lesion in the anterior epidural space from L3 to L4 and an abnormal signal in the L4 vertebral body. The patient underwent emergency surgery on the first day of admission for quick onset of paraplegia and dysuria. But the operation was stopped because of heavy bleeding. Five days later, she underwent endovascular embolization of the left and the right L4 lumbar segmental arteries, and after that, the hypervascular mass was subtotally removed. The histopathological diagnosis was choriocarcinoma. Then the patient received chemotherapy. But there was no apparent improvement in the neurological statement after chemotherapy. The patient's outcome is death.

**Conclusions:** This case suggests that choriocarcinoma should be borne in mind when observing a spinal extraosseous extension lesion with elevated  $\beta$ -HCG in a woman of child-bearing age.

## Background

Choriocarcinoma has known as a rare tumor and the most malignant type of gestational trophoblastic neoplasia (GTN). Gestational choriocarcinoma may be followed by hydatidiform moles, abortion, normal pregnancy, and ectopic pregnancy [1, 2]. It is characterized by the high secretion of human chorionic gonadotropin (HCG), rapid invasion of surrounding tissues, and early hematogenous metastasis. The common sites of metastasis outside the pelvic are lung, liver, kidney, and gastrointestinal tract [3-6]. The vertebrae metastasis of choriocarcinoma is extremely rare [4].

The present report describes a rare case of a middle-aged woman with metastatic choriocarcinoma in the lumbar vertebrae, epidural space, lung and adrenal gland which was admitted to The Second Affiliated Hospital of Dalian Medical University in January 2019.

## Case Presentation

A 42-year-old female patient was admitted to the spine surgery department with signs of low back pain for 10 days, weakness in both lower limbs and dysuria for 4 days. She gave a history of normal vaginal delivery of an infant 13 years ago and surgical treatment of chocolate cyst of the ovary 3 years ago. On neurophysical examination, the muscle strength was grade 0/5 for the left lower limb movement and grade 3/5 for the right. Deep tendon reflexes of both lower limbs were not elicited, and pathologic reflexes

were all negative. The patient also had decreased sensation below both knee joints. Sagittal and axial images using Magnetic resonance image (MRI) of the lumbar vertebrae revealed a nodular lesion in the anterior epidural space from L3 to L4 vertebral level with inhomogeneous low intensity on T1-weighted image (T1WI) and T2-weighted image (T2WI) (Fig. 1). There was also an abnormal signal in the L4 vertebral body with low intensity on T1WI and high intensity on T2WI (Fig. 1). A computed tomography (CT) scan of the chest showed about 4 cm sized block mass lesion in the right pulmonary hilar and multiple nodules in both lungs (Fig. 2). CT scans also showed tumors in bilateral adrenal glands and multiple retroperitoneal lymph nodes (Fig. 2). Transvaginal ultrasonography with Doppler and brain MRI were normal. Laboratory tests showed nothing significant except the elevated serum  $\beta$ -HCG level (9890 mIU/ml). Combined with some examinations, we initially considered that the lesions in the lumbar vertebrae and spinal canal might be a tumor or hematoma. At the same time, the patient's elevated HCG was most likely to be an ectopic pregnancy. So on the first day of admission, emergency surgery was performed due to the quick onset of paraplegia and dysuria. The spinous process, lamina, and ligament flavum of L3 and L4 were excised using a posterior approach. Profuse bleeding from the front wall of the spinal canal was challenging to control, and the operation was interrupted for hemostasis, after which posterior instrumentation using a pedicle screw and rod system was applied from L3 to L5. With the improvement of other auxiliary examinations (as shown above), we were inclined to diagnose the patient with a tumor, but the location of the primary lesion was uncertain. Five days after admission, she underwent angiography via a transfemoral approach with subsequent selective catheterization of the lumbar segmental artery, and this angiography revealed a hypervascular mass that was being supplied by the left and the right L4 lumbar segmental arteries. She then underwent a selective embolization (Fig. 3). On the same day, a second stage procedure of the surgery was performed. Following the previous path into the spinal canal, the epidural lesion appeared as a dark red encapsulated mass, which covered the cauda equina and the left L4 nerve root. The lesion was about 3 cm in diameter on the ventral surface of the dura, and half of which entered the subdural space through the rupture of the dural sac. Through the area lateral to the dural sac, the tumor was subtotally removed (Fig. 4). The pathological diagnosis was choriocarcinoma. Immunohistochemical staining of the biopsy revealed positivity for  $\beta$ -HCG, AE1/AE3, and Ki-67 (Fig. 5). She was then transferred to the oncology department, and her chemotherapy protocol was EMA/CO, which included etoposide, methotrexate, and actinomycin (EMA) plus cyclophosphamide and vincristine (CO). There was no apparent improvement in the neurological statement after chemotherapy. 8 months after surgery, the follow-up MRI showed abnormal signals in the S1 vertebrae, which is similar to the previous L4 vertebrae signal (Fig. 6), and her serum  $\beta$ -HCG level was still high (675 mIU/ml). Unfortunately, the patient was dead 11 months later after the surgery.

## Discussion

Choriocarcinoma is a rare malignant neoplasm that develops in the placental chorionic epithelial tissue niche. It is most commonly due to malignant transformation of hydatidiform moles, although it can develop following term pregnancy, spontaneous abortion, and even ectopic pregnancy [1]. It is known for a rapid hematogenous spread to multiple organs [3]. Approximately 30 % of cases of choriocarcinoma

have metastatic disease at the time of diagnosis. Lungs (80%) are the most common site of metastasis, followed by the vagina (30%), liver (10%), and brain (3-28%). However, skin, gastrointestinal tract, kidney, breast, and bones are infrequent sites [7-9]. The clinical signs of choriocarcinoma are so much varied that every case may be one of its kinds and thus can be a diagnostic challenge.

In our case, the patient presented with metastasis to the lung, lumbar vertebrae, epidural space, and adrenal gland. The lung has been described as the most common site for choriocarcinoma metastasis [7]. However, metastasis to the adrenal gland, lumbar vertebrae, and epidural space is extremely rare. Only 5 cases of metastatic choriocarcinoma in the lumbar and/ or epidural space have been reported thus far in the recent 20 years [3-5, 10, 11], and metastasis to adrenal gland has not been reported. Although we did not have any direct anatomopathological evidence related to the adrenal gland, image findings of abdominal CT and the reduction of the lesion after chemotherapy suggested that such metastases were present.

In our case, MRI of the lumbar vertebrae revealed a nodular lesion in the anterior epidural space from L3 to L4 vertebral level with heterogeneous low intensity on T1WI and T2WI. There was also an abnormal signal in the L4 vertebral body with low intensity on T1WI and high intensity on T2WI. The L4 vertebral body was not severely damaged, and the shape of the pedicle was standard. But its architecture of the cortical bone in the anterior epidural space was discontinuous, which can be seen on CT and MRI of lumbar vertebrae. Combined with these findings, our initial diagnosis was a tumor or hematoma, while the patient had an ectopic pregnancy. To quickly recover the muscle strength of the patient's lower limbs and improve symptoms of dysuria, surgery of the lumbar spinal canal decompression and mass resection was required. However, the profuse intraspinal bleeding interrupted the operation, which also confirmed the high possibility of malignancy because the bleeding could be caused by the rupture of blood vessels that feed malignant tumors.

Because of the affinity of trophoblast cells for blood vessels, metastases of choriocarcinoma often develop early and are mostly hematogenous. Due to the inherent ability of trophoblast cells to erode the walls of blood vessels and the fact that choriocarcinoma can pass through fragile blood vessel walls, the symptoms of metastases are frequently hemorrhagic. In our case, the patient had elevated  $\beta$ -HCG, but no pregnancy capsule was found in the uterus. We first considered the possibility of ectopic pregnancy but ignored the possibility of a trophoblast tumor. With the findings of tumors in both lungs and adrenal gland, we preferred to use metastatic carcinoma to explain all the results of the patient. Nonetheless, the exact location of the primary lesion was still unclear.

## Conclusion

We report here a sporadic case of lumbar vertebrae, epidural space, lung and adrenal gland metastases of choriocarcinoma. This case suggests that choriocarcinoma should be borne in mind when observing a spinal extraosseous extension lesion with elevated  $\beta$ -HCG in a woman of child-bearing age.

# List Of Abbreviations

HCG: human chorionic gonadotropin, GTN: gestational trophoblastic neoplasia, MRI: Magnetic resonance image, T1WI: T1-weighted image, T2WI: T2-weighted image, CT: computed tomography

## Declarations

### Ethical approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of institutional and/or national research committee.

### Consent for publication

The authors thank the patient for informed consent to this publication and the related images. All authors approved the final version to be published

### Availability of data and materials

The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

### Competing interests

The authors declare that they have no competing interest.

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### Authors' contributions

LL contributed to this manuscript: case management, literature research, and writing a major part of the manuscript. YZ contributed to this manuscript: editing of the manuscript and pathologic analysis. WZ contributed to this manuscript: case management, surgical procedure and manuscript revision. ZZ and ZL contributed to this manuscript: case management, surgical procedure and providing useful insights. All authors read and approved the final manuscript.

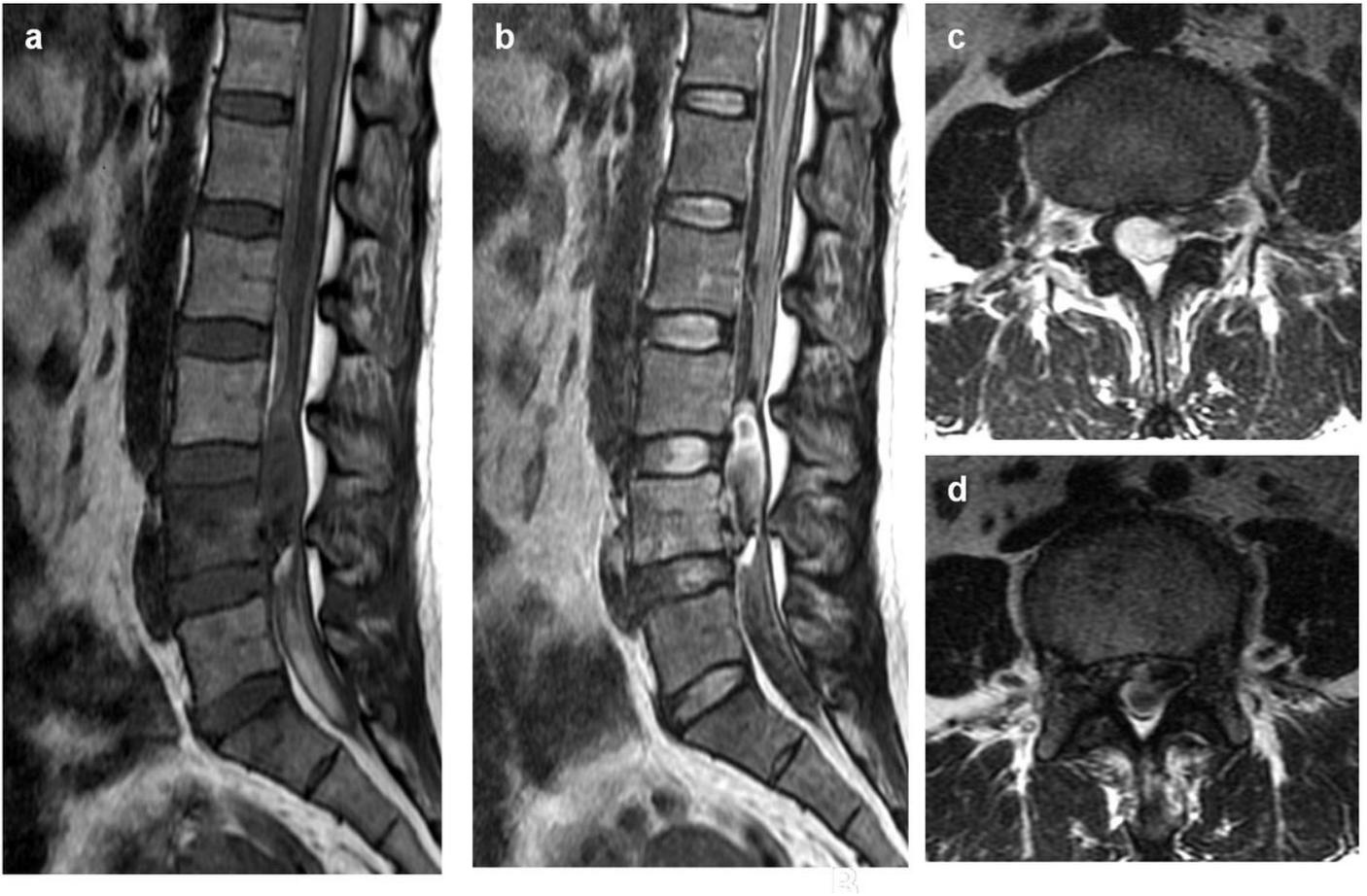
### Acknowledgements

The authors thank the patient for informed consent to this publication and the related images.

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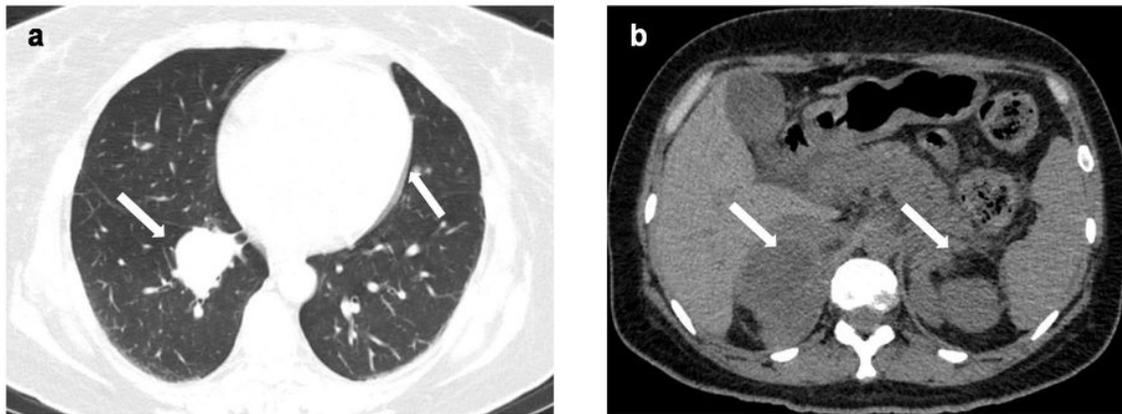
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## Figures



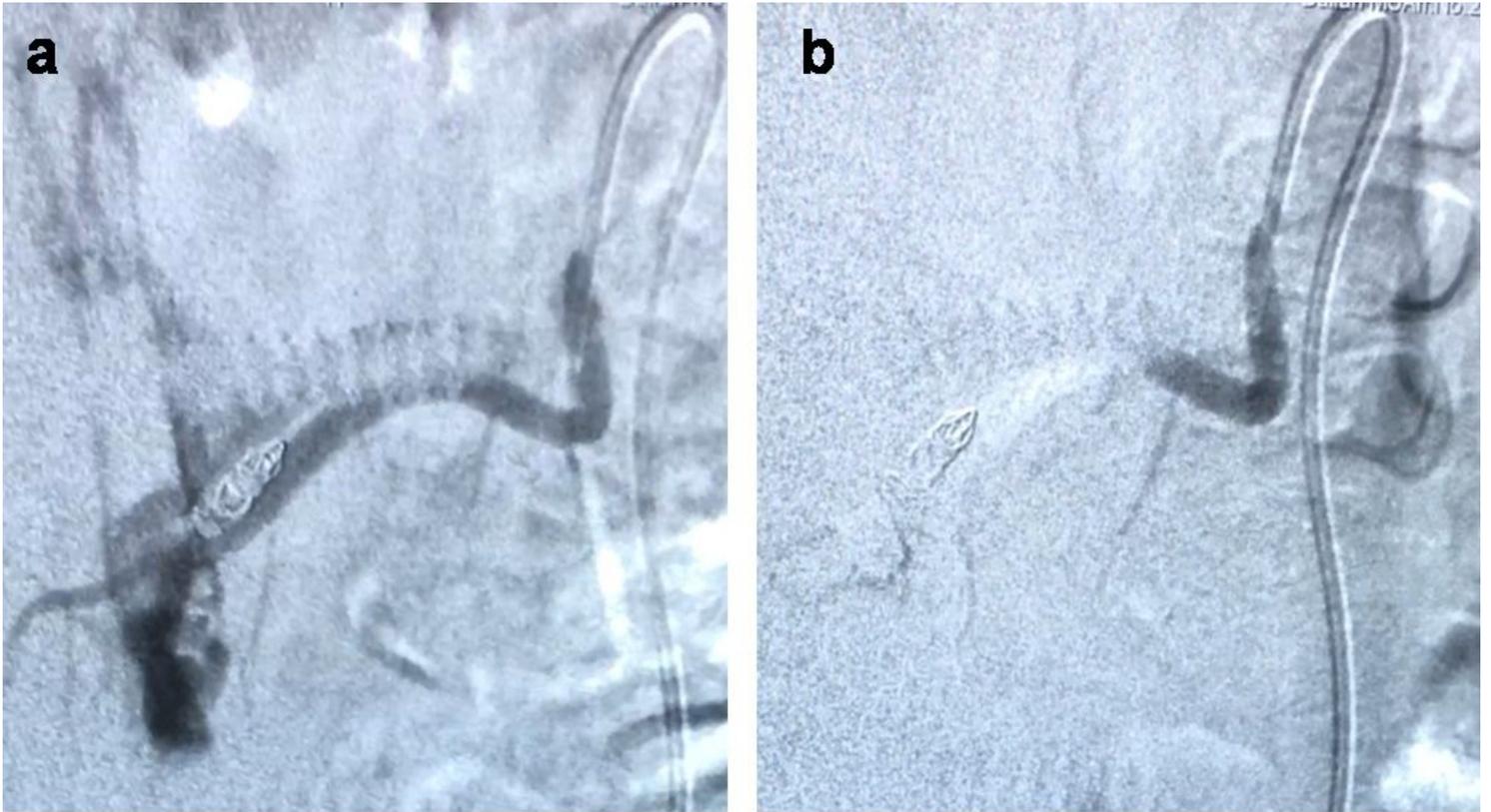
**Figure 1**

Preoperative magnetic resonance image of the lumbar vertebrae. Sagittal section of T1-weighted image (a) and T2-weighted image (b). Axial section of L3-4 vertebral space (c) and L4 vertebra (d).



**Figure 2**

Chest and abdominal computed tomography scans of the patient. Multiple metastases in both lungs (a) and bilateral adrenal glands (b). The arrows indicate the locations of tumors.



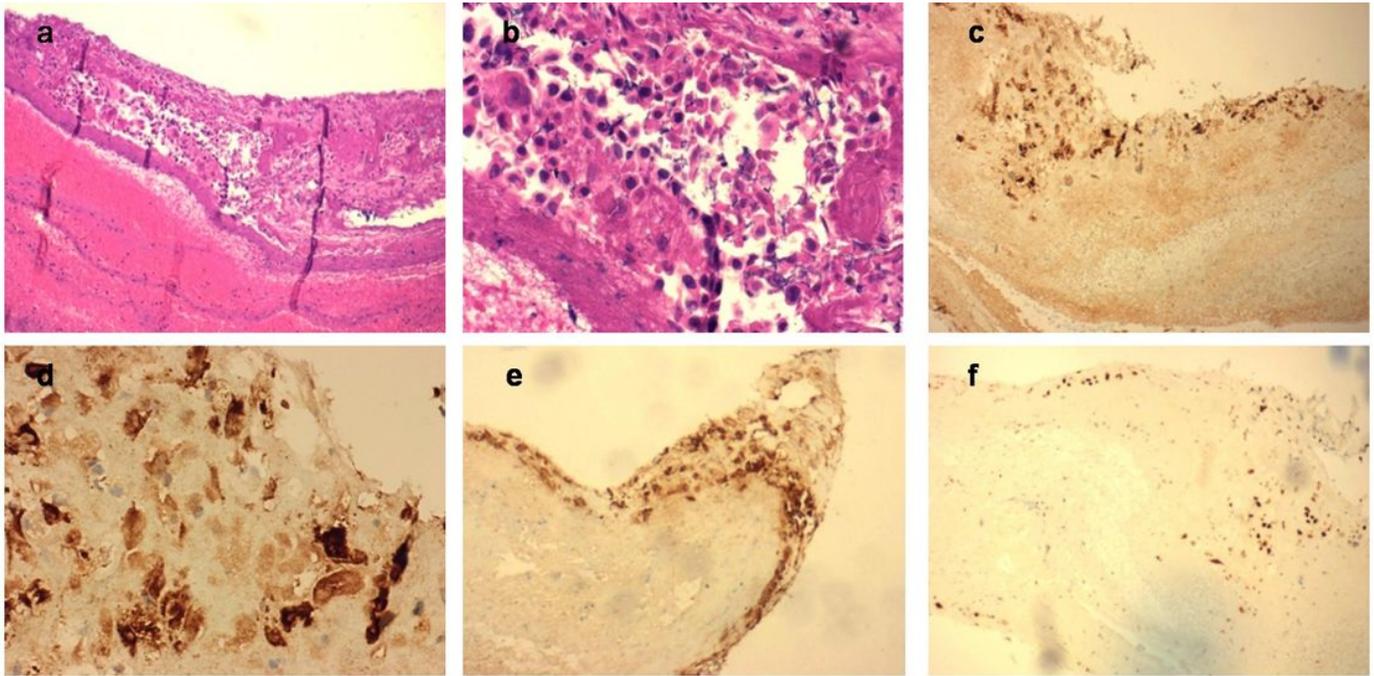
**Figure 3**

Selective angiograms of the left and right L4 lumbar segmental artery. (a) The pre-embolization angiogram shows a hypervascular mass supplied by the left and right L4 lumbar segmental arteries. (b) The post-embolization angiogram demonstrates the embolization of hypervascular mass with gelfoam and spring coil.



**Figure 4**

Pathological tissue. The tissue appeared as a dark red encapsulated mass and is about 3 cm in diameter.



**Figure 5**

Histopathological characteristics of the removed mass biopsy. There are cytotrophoblastic and syncytiotrophoblastic cells (H&E staining) without villi. (a) Magnification,  $\times 100$ . (b) Magnification,  $\times 400$ . Immunohistochemical staining of the biopsy revealed positivity for  $\beta$ -HCG (c, magnification,  $\times 100$ , d, magnification,  $\times 400$ ), AE1/AE3 (e, magnification,  $\times 100$ ), and Ki-67 (f, magnification,  $\times 100$ ).



**Figure 6**

Magnetic resonance image of the lumbar vertebrae at eight months after the operation. Abnormal signals are shown in the S1 vertebrae with low intensity on T1-weighted image (a) and high intensity on T2-weighted image (b).