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Case Report

A Case of Lumbar Metastasis of Choriocarcinoma Masquerading as an Extraosseous Extension of Vertebral Hemangioma

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We report here on an uncommon case of metastatic choriocarcinoma to the lung, brain and lumbar spine. A 33-year-old woman was admitted to the pulmonary department with headache, dyspnea and hemoptysis. There was a history of cesarean section due to intrauterine fetal death at 37-weeks gestation and this occurred 2 weeks before admission to the pulmonary department. The radiological studies revealed a nodular lung mass with hypervascularity in the left upper lobe and also a brain parenchymal lesion in the parietal lobe with marginal bleeding and surrounding edema. She underwent embolization for the lung lesion, which was suspected to be an arteriovenous malformation according to the pulmonary arteriogram. Approximately 10 days after discharge from the pulmonary department, she was readmitted due to back pain and progressive paraparesis. The neuroradiological studies revealed a hypervascular tumor occupying the entire L3 vertebral body and pedicle, and the tumor extended to the epidural area. She underwent embolization of the hypervascular lesion of the lumbar spine, and after which injection of polymethylmethacrylate in the L3 vertebral body, total laminectomy of L3, subtotal removal of the epidural mass and screw fixation of L2 and L4 were performed. The result of biopsy was a choriocarcinoma.

KEY WORDS : Metastatic choriocarconoma · Spinal metastasis.

INTRODUCTION

Choriocarcinoma is a rare tumor and it is the most malignant and aggressive neoplasm of all the gestational trophoblastic diseases (GTDs). It can appear after an intrauterine or an ectopic pregnancy and it is more common after a hydatidiform mole. Choriocarcinoma is associated with a high human chorionic gonadotrophin (HCG) level and rapid hematogenous spread to multiple organs[1,7,8,12,20,22,23,25]. The most common locations for metastases are the lung and the vulvo-vaginal region, and metastases are less commonly found in the brain and liver. Other sites of metastases, such as the skin, gastrointestinal tract, kidney, breast or bones, are extremely rare[2,4,11,12,15,20-24]. In this study, we report on a case of metastatic choriocarcinoma that presented with intracerebral hemorrhage and pulmonary hemorrhage and it metastasized to the lumbar spine, and this metastasis to the spine radiologically mimicked an extraosseous extension of vertebral hemangioma.

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CASE REPORT

A 33-year-old female patient was admitted to the pulmonary department with headache, dyspnea and hemoptysis. Her medical history included a cesarean section due to intrauterine fetal death at 37-weeks gestation and this occurred 2 weeks before admission to the pulmonary department. The patient had no abnormal findings on physical examination. However, a computed tomography (CT) scan of the chest showed about a 2 cm sized nodular mass lesion and hemorrhage in the left upper lobe of the lung with hypervascularity, and this was all suspected to be an arteriovenous malformation (AVM). On CT of the brain, a hemorrhagic lesion measuring 22 mm in size was seen with a focal, highly attenuated, subtle, peripheral enhancing

nodule with peripheral edema in the right parietal lobe, which was consistent with a vascular lesion, but a magnetic resonance (MR) angiogram of the brain did not reveal a definite vascular lesion (Fig. 1). She successfully underwent

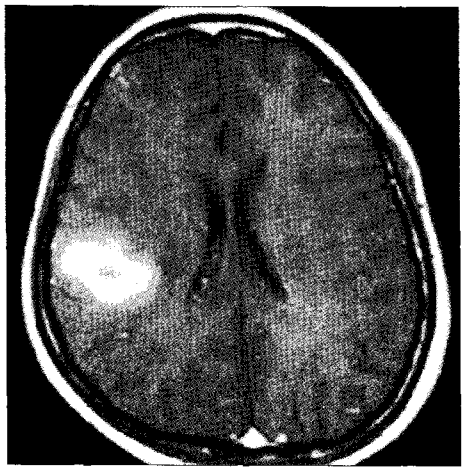

Fig. 1. Brain magnetic resonance image (T2WI) : A hemorrhagic lesion measuring 22 mm in size is seen with a focal, highly attenuated nodule with peripheral edema in the right parietal lobe.

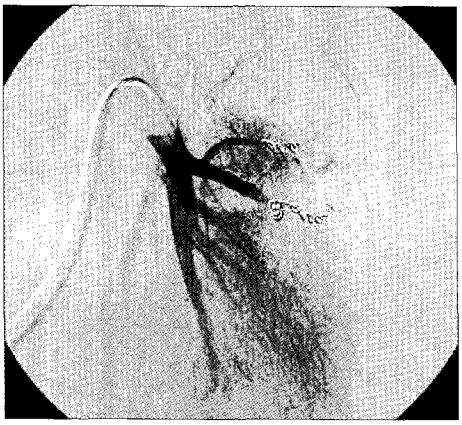

Fig. 2. Pulmonary angiograph : The post-embolization angiogram demonstrating the embolization of the pulmonary hypervascular lesion with microcoils.

embolization of the left pulmonary hypervascular lesion with microcoils (Fig. 2). The symptoms leading to admission subsided and a second brain CT revealed that the hematoma was more attenuated. She was discharged 3 weeks after admission and she was scheduled for transfemoral cerebral angiography.

Approximately 10 days after discharge from the pulmonary department, she was readmitted to the due to back pain and a sudden onset of weakness in both lower limbs. On neurological examination, the limb power was grade IV for the ankle and grade III for the knee movement. The deep tendon reflexes in both legs were normoflexic and there was a negative Babinski sign. A plain radiograph of the lumbar spine was normal, yet the MR image of the lumbar vertebrae revealed a low signal intensity lesion on the T1-weighted images (T1WI) and a high signal intensity lesion on the T2-weighted images (T2WI) affecting the entire L3 vertebral body with the pedicle and extending to the epidural space in a curtain shape, and the lesion was well enhanced (Fig. 3). But, on the CT and MR image of the lumbar spine, the trabeculation pattern of the L3 vertebra was preserved and the architecture of the cortical bone of the L3 vertebra was also intact (Fig. 4). According to these findings, we considered this lesion to be an extraosseous extension of vertebral hemangioma.

She first 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 from the L3 lumbar segmental artery on the right (Fig. 5). She then underwent a selective embolization using polyvinyl acetate (PVAc) particles. On the first day after embolization, she was administered a percutaneous injection of ethanol into the L3 vertebral body as a sclerosing agent to rapidly obliterate and shrink the mass[3,5,9,10]. We used a bipedicular approach with

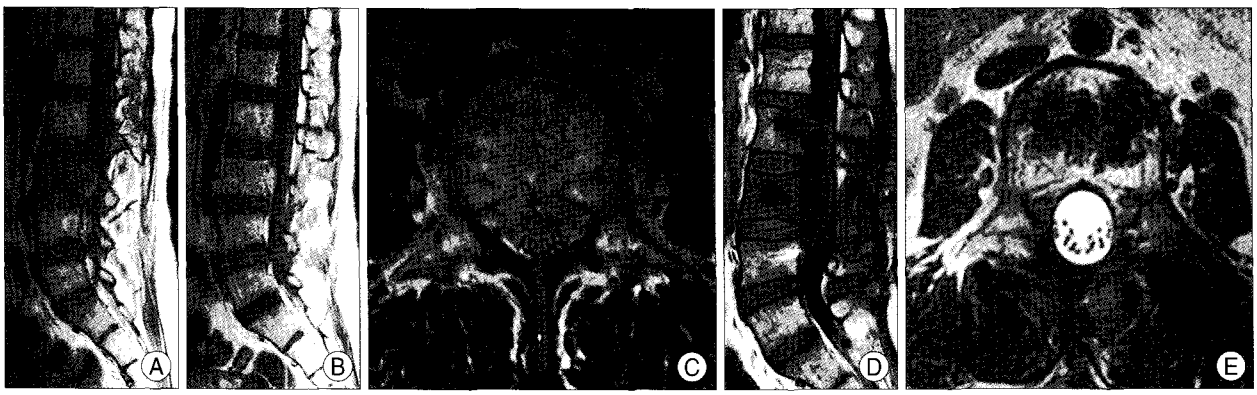

Fig. 3. Pre-and post-operative magnetic resonance images. The preoperative magnetic resonance images of the lumbar spine showing low signal intensity on T1WI (A) and focal enhancement on T1C+ (T1-weighted image enhanced) (B), and a lesion affecting the entire L3 vertebral body with the pedicle and extending to the epidural space in a curtain shape (C). The postoperative magnetic resonance images showing a significantly decreased epidural lesion compared with the preoperative images (D and E).

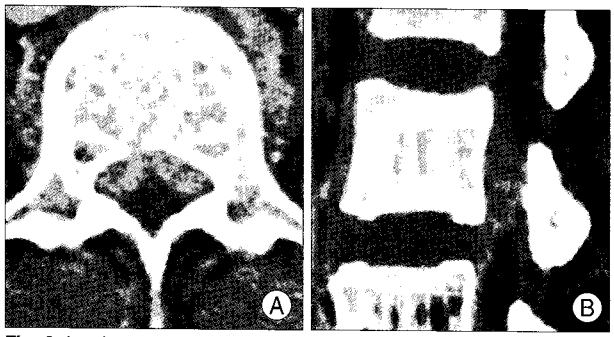

Fig. 3. Lumbar computed tomography (CT) scan of the L3 vertebra. The axial CT scan (A) and sagittal reconstruction CT scan (B) showing the vertical coarse bony trabeculae and soft tissue mass extending to the epidural space in a curtain shape, but the architecture of the cortical bone of the L3 vertebra is intact.

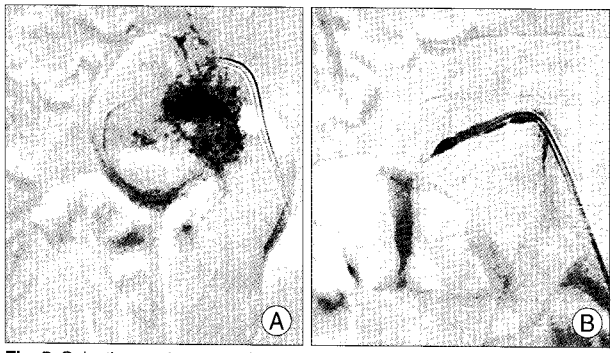

Fig. 5. Selective angiograms of the right L3 lumbar segmental artery. A : The pre-embolization angiogram showing a hypervascular lesion supplied from the right L3 segmental artery. B : The post-embolization angiogram demonstrating the embolization of the hypervascular lesion with polyvinyl acetate.

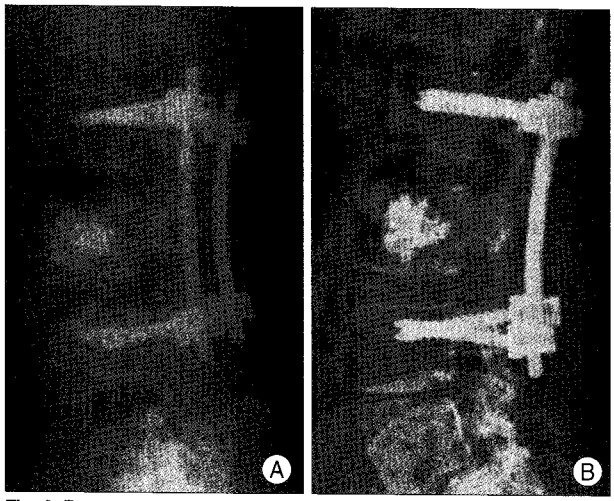

Fig. 6. Postoperative radiographs : X-ray on the first day after the operation (A) showing the injected PMMA cement on the L3 vertebral body, total laminectomy of L3, and L2-4 pedicle screw fixation. A follow-up X-ray ten months later (B) doesn't show any further bony destruction.

4 ml of absolute ethanol in each pedicle. Yet, the weakness in both lower extremities progressed rapidly immediately after the ethanol injection, so posterior decompression and stabilization were performed. The lesion was approached through a wide laminectomy of L3 and bilateral foraminotomy at L3-4 (Fig. 6). The epidural lesion appeared as a dark red encapsulated mass. The lesion was moderately adhered to the dural sac. Under the surgical microscope, the epidural tumor was subtotally removed through the space lateral to the thecal sac. A Jamshidi needle was inserted into the L3 pedicle bilaterally and about 6 mL of polymethylmethacrylate cement was injected under fluoroscopic guidance. After this, L2-4 pedicle screw fixation was performed. She remarkably recovered from her paraplegic state to grade IV. The histopathological diagnosis was a choriocarcinoma (Fig. 7). Immunohistochemical staining of the biopsy revealed positivity for pancytokeratin, beta-human chorionic gonadotropin (β-HCG) and human placental lactogen. Her postoperative β-HCG level in serum was above 100,000 mIU/mL. A postoperative CT scan of the abdominopelvic area revealed an enlarged uterus with heterogeneous enhancement. She was then transferred to the hemato-oncology department for chemotherapy. She received combination chemotherapy in the form of EMA-CO, which included etoposide, methotrexite and actinomycin (EMA) plus cyclophosphamide and vincristine (CO). There was improvement of the neurological status with chemotherapy, and she became able to walk around. Ten months after operation, the follow-up MR images showed a significant decrease in the size of the epidural lesion and an intact bony contour (Fig. 3) and her β-HCG level in the serum returned to normal (1 mIU/mL). She is currently being regularly monitored in the gynecology department.

DISCUSSION

GTDs encompass a heterogeneous group of neoplastic disorders that arise from the trophoblastic epithelium of the placenta and they are characterized by a distinct tumor marker (β-HCG)[1,7,8,11,12,17,20,22,23,25]. GTDs are conventionally classified into at least five distinct groups on the basis of their histopathologic, cytogenetic, and clinical features[12]. They are complete and partial hydatidiform mole, invasive mole, choriocarcinoma, placental site trophoblastic tumor and miscellaneous trophoblastic lesions. Choriocarcinoma is a rare, highly malignant neoplasm of a trophoblastic origin among the GTDs. This tumor is known for its association with molar pregnancy, a rapid hematogenous spread to multiple organs, high HCG levels and a good response to chemotherapy[1,12,17,19,22]. It is preceded by several conditions as follows : 50% arise in molar pregnancies, 25% arise after previous abortions, 23% arise in normal pregnancies and 3% arise subsequent to ectopic pregnancies[11].

Choriocarcinoma has a marked tendency to metastasize early by blood-borne dissemination, like in our case. Wides-

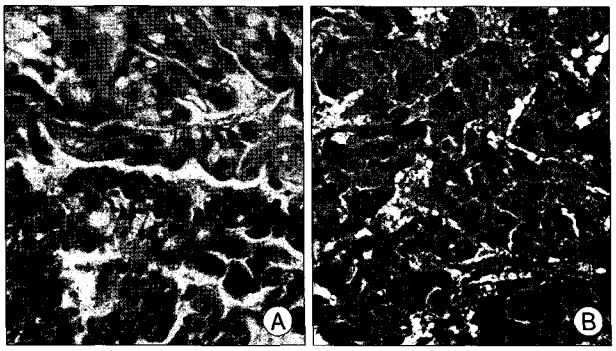

Fig. 7. Histological findings of the removed mass. A : There is a mixture of cytotrophoblastic and syncytiotrophoblastic cells (H & E, ×200). B : Human chorionic gonadotrophin positive cells are observed immunohistochemically (×200).

pread metastases are characteristic of choriocarcinoam. The favored sites of involvement are the lungs (94% of all metastatic choriocarcinoma), vagina (44%), liver (28%) and brain (28%), followed by the skin, gastrointestinal tract, kidney, breast and bones[11,24]. The clinical signs can be very different, depending on the site of the lesions. The disease often presents with symptoms related to metastatic spread as the primary tumor may remain very small[11,22].

Approximately 30% of the patients with choriocarcinoma show metastases at the time of diagnosis[20]. In the case we have described, the patient presented with metastases to the lung, brain parenchyma and lumbar spine. The lung and brain have been described as the most common sites for metastasis in the literature[1,2,4,11,12,15,20,21,23,25]. Yet, metastasis in the musculoskeletal system, such as the spine, is extremely rare and this can be seen from the scant reported cases[4,14-16,20,21]. This current report presents an unusual metastatic choriocarcinoma in the lumbar spine and it extended to the epidural space.

MR image of the lumbar vertebrae in our case revealed a low signal intensity lesion on T1WI and a high signal intensity lesion on T2WI, and this lesion affected the entire L3 vertebral body with the pedicle; the lesion extended to the epidural space with a curtain shape and it was well enhanced. But, on CT and MR image of the lumbar spine, the trabeculation pattern of the L3 vertebra was preserved and the architecture of the cortical bone of the L3 vertebra also is intact. With these findings, our initial impression was an extraosseous extension of vertebral hemangioma that was aggravated during pregnancy and the puerperal period. Pregnancy is a known risk factor for symptomatic conversion of a vertebral hemangioma[6,13]. It is also widely believed that pregnancy and the puerperium are associated with an increased risk of hemorrhage and the aggressive behavior of carvernous malformations[20].

Beskonakli et al.[4] reported on a 44-year-old woman with metastatic choriocarcinoma of the thoracic spine that extended to the extradural space and this caused paraplegia. Myelography showed a complete block with intact bony structures at the T5 level. Menegaz et al.[15] reported on a 45-year-old woman with metastatic choriocarcinoma to the lumbar and sacral segments. The radiologic findings showed a predominantly extrathecal intracanalicular process extending from L2 to S1 and compressing the thecal-radicular structures. But, the bony structure that was involved was preserved and it exhibited a high signal in T2WI, which suggests microfractures or reactional hyperemia. These findings suggest the choriocarcinoma metastasizing to bony structure has a tendency to invade ahead to the soft tissue and later destroy the bony elements.

Metastases often develop early and they are generally hematogenous because of the affinity of trophoblasts for blood vessels. Because the reported choriocarcinomas were often perfused by fragile vessels, as well as the innate capacity of trophoblastic cells to invade and erode vessel wall, they were frequently hemorrhagic[11,24]. The symptoms of metastases usually result from bleeding at metastic foci. Bony metastases secondary to choriocarcinoma are exceptional. Nonetheless, because the lung and brain are the most common sites of metastasis, many articles[1,2,4,11,12,15,20-23,25] have emphasized that the possibility of choriocarcinoma should be borne in mind when observing parenchymal hemorrhage of the lung or any intracranial bleed in woman of child-bearing age. In our case, the pulmonary and cerebral hemorrhagic content existed and furthermore, a spinal hypervascular mass with feeder vessel was seen on arteriography. As the tumor occupied the L3 vertebral body and this was combined with spinal vasculopathy, which mimicked vertebral hemangioma, we mistook it for a vertebral hemangioma that was aggravated during the puerperal period. Additionally, we missed checking the serum β-HCG level on the patient's laboratory tests. Determining the β-HCG level is necessary for making the diagnosis of choriocarcinoma, and is also useful for the follow-up to detect recurrence and as a prognostic marker[1,7,8,11,12,20,22,23,25]. Choriocarcinoma is characterized by causing autonomous secretion of β-HCG.

CONCLUSION

We report here on an uncommon case of metastatic choriocarcinoma to the lung, brain and lumbar spine. When a spinal extraosseous extension lesion like a spinal hemangioma which aggravated during pregnancy or the puerperal period is encountered, the possibility of metastatic gynaecological disease such as choriocarcinoma should be borne

in mind, and especially in fertile female patients.

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