

Case Report

Metastatic choriocarcinoma to the lumbar spine: Case report and review of literature

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Abstract

Background: There are few cases of choriocarcinoma metastases to the spine that have been reported. Most occurrences are in women with the gestational form of the tumor, and these now exhibit a very high remission rate with chemotherapeutic treatment, typically circumventing the need for spinal surgery.

Case Description: In an effort to better understand treatment options for those rare instances when choriocarcinoma does find its way into the spine, we have synthesized a comprehensive literature review on the clinical cases of choriocarcinoma spinal metastases. We also describe our unique experience and decision-making involving the first reported case of surgical treatment of non-gestational choriocarcinoma spinal metastases in a male patient.

Conclusion: Spinal surgery has a limited role in metastatic choriocarcinoma, but there is the potential for improving neurologic decline even in the rare and aggressive male variant of this disease.

Key Words: Cauda equina, chemotherapy, choriocarcinoma, germinoma, tumor

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INTRODUCTION

Choriocarcinoma is a germ cell cancer that occurs in two distinct forms, a *de novo* non-gestational form and, more commonly, a gestational form that consists of abnormal trophoblastic cell growth. The latter type is unique in that it occurs in female hosts, but the tumor cells contain foreign genetic material of spermatid origin. While both forms were considered deadly neoplastic diseases at one time, choriocarcinoma has become known as one of the great success stories in cancer chemotherapy. Women suffering from even advanced stages of gestational choriocarcinoma can now expect a good response or cure from appropriate

chemotherapeutic regimens. However, non-gestational choriocarcinoma, while very rare, remains a very fatal disease process that remains largely unresponsive to current treatment efforts.

Choriocarcinoma occurring in a male patient always represents the non-gestational form. There are very few reports of metastases to the spine, and even fewer are described in males. Here, we present the most comprehensive cross-disciplinary literature review to date of cases that include any description of choriocarcinoma metastases to the spine either pre- or post-mortem. Also, we present the first description of spinal surgery on a male patient with metastatic choriocarcinoma.

CASE REPORT

A 30-year-old male presented to an outside facility with the complaint of increasing low back pain over the past month that radiated bilaterally to the level of his ankles. The pain was accompanied by progressive lower extremity weakness to the point where ambulation became impossible 3 days prior. He also complained of bilateral calf and anterior thigh numbness with paresthesias. He denied changes in normal bowel and bladder function. He had been evaluated 3 weeks prior at the same facility with similar but less severe complaints and diagnosed with bilateral sciatica. Workup during the more recent visit revealed an L2 mass invading the spinal canal for which the patient was transferred to our facility for neurosurgical evaluation [Figure 1].

The patient had noted a left testicular mass 3 years ago and stated that it had been enlarging in size and was painful to touch. He denied having ever brought this complaint to the attention of a physician. He also admitted to poor appetite and significant weight loss over the prior month amounting to 25 pounds and attributed this to his major depression. He denied fevers, vision changes, night sweats, or any other palpable lesions or rashes.

Physical examination

The patient was a pale, anxious-appearing male who was alert and cooperative, tachycardic, and hypertensive. He exhibited tenderness to palpation in the midline lumbar spine. Upper extremity strength was normal. Right hip flexion, knee extension, dorsiflexion, and plantar flexion were grade 4/5. The same motor groups were only at anti-gravity strength (3/5) on the left side. Rectal sphincter function was normal. On sensory exam,

saddle anesthesia was noted as was decreased sensation to light touch in a distribution that approximated the bilateral L2 and L3 dermatomes. His reflexes were grade 2+ throughout and no long-track signs were observed. His left testicle was enlarged and a mass was palpable that was non-tender at the time.

Review of diagnostic studies

A magnetic resonance image (MRI) of the spinal axis demonstrated a 12 cm left-sided retroperitoneal mass that extended into the spinal canal at the L2 and L3 levels through the left L2 and L3 neural foramen with severe thecal sac compression at the L2 level [Figure 1]. A scrotal ultrasound showed an echogenic region within the left testis with clustered microlithiasis suspicious for primary testicular neoplasm. Initial serum labs were remarkable showing the following levels: Sodium 131 mmol/l, alkaline phosphatase 291 IU/l, hemoglobin 6.7 g/dl with serum iron level being 13 µg/dl and ferritin level of 2294 ng/ml, albumin 2.3 g/dl, sedimentation rate 125 mm/h, and β-Human Chorionic Gonadotropin 248,379 IU/l.

Operation and postoperative course

In the first 24 h after admission, during his inpatient medical workup, the patient's lower extremity motor exam deteriorated rapidly to less than anti-gravity bilaterally. A decision was made to urgently decompress and stabilize the spine, as well as obtain tissue diagnosis before the full metastatic workup had been completed. Bilateral laminectomies were performed from L1 through L3. Soft tumor mass was removed extradurally from within the canal, resulting in thorough decompression of the conus and cauda equina. Surprisingly, blood loss was not particularly bad and transfusion was not needed. Frozen intraoperative pathology was reported

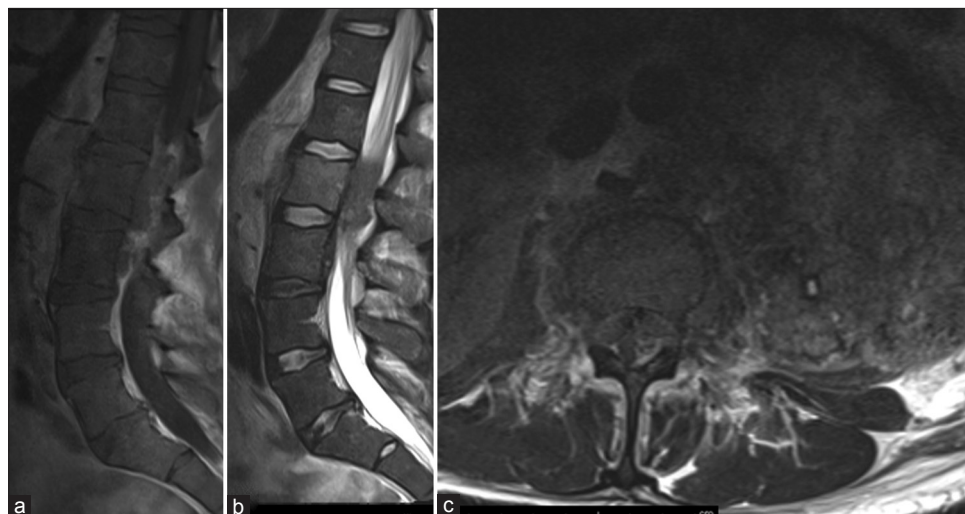


Figure 1: (a) Sagittal T1-weighted enhanced image showing heterogeneously enhancing epidural mass centered at L2. (b) Sagittal T2-weighted image illustrates compressive mass surrounding the cauda equina. (c) Axial T1-weighted enhanced image shows massive left retroperitoneal lesion extending into the L2 neural foramen and epidural space

as metastatic carcinoma. As the amount of bone and tumor removed was likely destabilizing and the bone quality found to be poor at these levels, a posterolateral pedicle screw fusion was performed from T12 through L4 [Figure 2].

Final histopathologic analyses revealed abundant tissue necrosis with an estimated live tumor cell presence of only 5-10% [Figure 3]. Immunohistological analysis indicated a final diagnosis of metastatic choriocarcinoma.

Postoperative metastatic disease burden workup revealed that the retroperitoneal mass was actually 13 × 14 × 26 cm and encased the aorta, splenic artery, the left renal artery, and vein. Also, there was extensive metastatic burden within both lungs and the liver. Free fluid was present within the pelvis. A contrasted MRI of the entire neuraxis revealed a hemorrhagic right occipital enhancing lesion, a non-enhancing subcortical left inferior frontal lobe lesion, and an enhancing C3 vertebral body lesion.

Within 24 h after decompression, the patient's bilateral lower extremity strength and sensation improved. He was maintained on dexamethasone for cerebral and spinal edema. Subcutaneous enoxaparin and sequential compression devices were used to prevent thromboembolism. Chemotherapy was initiated on postoperative day 9 and consisted of etoposide, ifosfamide, and cisplatin. On the same day that chemotherapy was started, his hospital course was complicated by acute respiratory failure secondary to a suspected pulmonary embolism. He was intubated and an inferior vena cava filter was placed to prevent additional emboli. He declined while intubated, requiring pressor support, and developed a large pericardial effusion which was drained via pericardial window. He was started on continuous renal replacement therapy, and his spouse elected to withdraw care compassionately on postoperative day 21 (hospital day 22) which resulted in death on the same day.

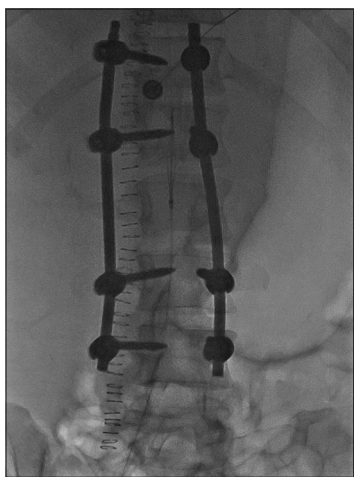


Figure 2: Postoperative radiograph demonstrating the fusion construct for stabilization spanning T12-L4

DISCUSSION

Choriocarcinoma is a rarely encountered lesion within the spine. Review of the modern English literature revealed only six descriptions of neurosurgical management of spinal metastases.^[2-4,10,11,14] The case described here is unique as the first case of surgical resection of a pure choriocarcinoma metastasis from the spine of a male patient. Table 1 presents all cases of reported spinal metastases found in a comprehensive literature search.^[1-4,6,9-11,13,14,16,21,22] The first three reported cases of spinal choriocarcinoma in other young males all represent cases of advanced multi-system disease burden as in the case reported here. In fact, in two of the three prior cases, the spinal involvement was only found upon autopsy.^[1,22] In the living male patient with spine, brain, kidney, lung, liver, and eye involvement, chemotherapeutic and radiation treatments have provided an excellent outcome for at least 4 years.^[6] With an incidence of 0.5% of all testicular tumors, pure choriocarcinoma is very rare in men and typically originates from totipotent gonadal germ cells but often presents with advanced metastases.^[15] Contrary to the success observed with chemotherapeutic treatment of choriocarcinoma of paternal genetic origin more commonly seen in women who have experienced a hydatidiform mole (80% remission even in widely metastatic cases), the prognosis in men with this disease remains grim.^[5,17] Of note, there are several cases reported with spinal metastases containing mixed germ cell tumors with choriocarcinoma elements; these mixed tumors, however, do not necessarily bear a worse prognosis on account of the choriocarcinoma mixture.^[12,17,19]

The decision to proceed with spinal decompressive surgery in this case was made on an urgent basis based on progressive critical neurologic deficit with radiographic evidence of severe impingement of the cauda equina. Choriocarcinoma metastases to the spine are exceedingly rare. Lesions are most commonly found in lung and brain in men, but as seen in Table 1, there is a propensity for epidural invasion and associated neurologic deficits when the spine is involved.^[18] However, in the absence of acute progressive neurologic decline or evidence of considerable instability, caution should be exercised in

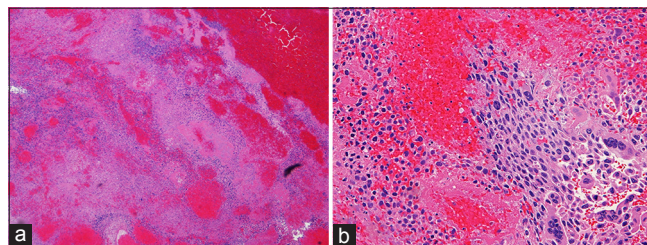


Figure 3: Hematoxylin and eosin pathologic stain of epidural tumor. (a) Low-power view showing diffuse hemorrhages common in choriocarcinoma. (b) High-power view highlighting syncytiotrophoblasts and large, pleomorphic malignant single cells

Table 1: Summary of case reports describing choriocarcinoma metastases to the spine in the medical literature

Primary author/Year	Age/ gender	Tumor location	Neurologic deficit	Spinal surgery	Chemotherapy	Radiation	Outcome
Azzopardi, 1961	26/M	Vertebrae (autopsy), retroperitoneal, liver, lung, brain, kidney, spleen, psoas, testis	None	No	Stilbestrol	No	Death 6 weeks from admission
Balat, 2004	24/F	T5 body, T3-5 epidural, ovary, sternum	Paresthesias	T5 corpectomy	BEP	No	Death after first chemotherapy cycle
Beşkonakli, 1998	44/F	T5 epidural, uterus	Difficulty ambulating, urinary incontinence	T4-T6 laminectomies, gross total removal	Not described	No	No neurologic improvement. Death 5 months after surgery
Eskreis, 1988	33/F	T2-3 epidural, stomach	L2 sensory loss, lower extremity weakness, urinary incontinence	T2-3 laminectomies	Methotrexate, actinomycin D, cytoxan	Lumbar spine	Full strength and continence resumed, alive at 6 weeks post-op
Guber, 2011	26/M	Spine, eye, lung, brain, kidney, liver	Visual acuity	No	BEP	Head, spine, eyes	Remission at 4 years follow-up
Ko, 2012	21/F	L2 body, thoracic intramedullary, brain, lungs	Altered mentation, field cut, paraplegia with T10 sensory level	No	Multi-agent	Thoracic and lumbar spine	Death 13 months after diagnosis
Kuten, 1978	20/F	L1-L3 epidural	Bilateral paraparesis	L1-3 laminectomies, partial resection	Methotrexate	Lumbar spine	Residual foot drop, no evidence of disease at 4-year follow-up
Lee, 2010	33/F	L3 body and pedicle with epidural extension, brain, lung, uterus	Bilateral paraparesis	Embolization, L3 laminectomy, L2-4 posterolateral fusion, L3 vertebroplasty	EMA-CO	No	Shrinking epidural mass, ambulatory, and normal β -HCG at 10 months
Menegaz, 2004	45/F	L2-S1 epidural, iliopsoas, lungs, uterus	RLE plegia, LLE paresis, paresthesias, urinary incontinence	No	EMA-CO	Lumbosacral	Death at 5 months from diagnosis
Natio, 2009	38/F	L2 body, lung	None	L2 vertebrectomy, L1-3 posterolateral fusion	Methotrexate	Lumbar	Death 3 months post-op
Rustin, 1989	F	Lumbar spine	Unknown	Unknown	EMA-CO	No	Unknown
Vani, 1993	27/F	S5, lung, gluteal	None	No	Methotrexate, actinomycin-D, chlorambucil, etoposide	Sacrum	Lost to follow-up
Williamson, 1994	26/M	Vertebral bodies (autopsy), testis, liver, spleen, kidneys, pancreas, thyroid, adrenals, eyes, lung	Visual acuity	No	Yes	Orbits	Death 1 month from diagnosis

M: Male, F: Female, BEP: Bleomycin, etoposide, EMA-CO: Etoposide, methotrexate and actinomycin, cyclophosphamide, RLE: Right lower extremity, LLE: Left lower extremity, HCG: Human chorionic gonadotropin

surgical decision-making. Choriocarcinoma tends to be a hypervascular lesion prone to hemorrhages; even radical spondylectomy has demonstrated failure of disease control.^[14,15] Given the potential for good response to radiation and chemotherapy, prophylactic or debulking surgery is not recommended. Palliative surgery should typically be reserved for scenarios where aggressive conservative management and trials of radiation and chemotherapy have failed to reduce symptoms. Emergency surgery for severe rapid-onset neurologic

decline due to compression should always be considered with disclosure that the surgical risks may be elevated because of bleeding or complications related to systemic tumor burden.

Testicular germ cell tumors are the most common neoplasms found in men in the 15-34 year age group with a “remarkably distinct distribution,” with the peak age being 30-34 years.^[7] The three previous cases of men with choriocarcinoma to the spine were all aged 26, and the patient described here would have been 26 as well when

he first noticed his testicular lesion. Therefore, serum markers for germ cell tumors should be sent as part of the initial workup for men in this age range presenting with metastatic disease. A simple urine HCG test can serve as a rapid indicator of choriocarcinoma (pure or mixed).^[20]

CONCLUSIONS

Metastatic choriocarcinoma to the spine of non-gestational trophoblastic origin is exceedingly rare and most likely represents end-stage disease. Spinal surgery has a limited role in this form of the disease, but could be considered for progressive neurologic decline. The success of chemotherapeutic treatment for gestational choriocarcinoma in women has not translated to other forms of this cancer in men. Nonetheless, select case reports describing surprising success in several men with advanced disease burden hold some hope for the possibility of medical cure.^[6,8]

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