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Case Report

Metastatic intradural extramedullary spinal cord tumor from ovarian cancer: A case report with a literature review

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Context: Metastatic intradural extramedullary spinal cord tumors are extremely rare.

Findings: A 76-year-old woman presented with intractable neck pain. Three years earlier, she had been treated for ovarian cancer with bilateral salpingo-oophorectomy. A year later, she underwent resection of a brain metastasis. Magnetic resonance imaging (MRI) showed an encapsulated intradural extramedullary mass at C4–C5. C4–C5 hemilaminectomy, tumor resection, and biopsy were performed. Histological examination of the resection revealed an adenocarcinoma. After surgery, her intolerable neck-shoulder pain was fully resolved, and she had no difficulties with daily living activities. However, two months later, she underwent gamma knife radiosurgery for the recurrent metastatic brain tumor, and four months later, she died from cachexia.

Conclusion: Although cases of metastatic intradural extramedullary spinal tumors from ovarian cancer are extremely rare, their possibility should be considered in the differential diagnosis. A history of brain metastases and enhancement on T1-weighted MRI were helpful for making an accurate diagnosis.

Keywords: Cervical spine, Spinal cord tumor, Metastasis, Ovarian cancer

Introduction

Ovarian cancer is the fifth leading cause of mortality among US women.¹ The reason for the high death rate is the late presentation in most cases, meaning that the disease is widely metastatic within the abdomen at the time of diagnosis. However, distant metastases seldom occur in patients with ovarian cancer, and ovarian cancer patients developing a central nervous system metastasis are exceptionally rare.² Only 5 out of 255 patients with ovarian cancer developed a central nervous system metastasis (1.95%), and all five patients had brain metastases.³

Metastatic epidural spinal compression is a common malignancy complication that affects almost 5% of patients with cancer.⁴ However, intradural metastases

of a non-neurogenic origin present an extremely rare manifestation of systemic cancer.⁵ Intradural extramedullary spinal cord metastasis is seen at autopsy in fewer than 5% of patients who have died from cancer.⁶

This report describes the case of a 76-year-old woman with ovarian cancer who presented with neck-shoulder pain. Intradural extramedullary spinal cord metastasis of ovarian cancer was confirmed by histopathologic examination. This report is the third report of ovarian cancer metastasized to the intradural extramedullary spinal cord in the literature.^{7,8} Clinical features of three cases, our case and two reported cases, are discussed and summarized for accurate preoperative diagnosis of this rare presentation.

Case report

A 76-year-old woman presented with 6 months of intractable neck pain. Though she was prescribed an opioid pain reliever, she reported excruciating pain in her neck that had awakened her from sleep for four

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months. Three years earlier she had been treated for ovarian cancer with bilateral salpingo-oophorectomy followed by chemotherapy. In addition, a year later she was treated for metastasis of the brain with a tumor resection. On examination, a Jackson test induced radicular pain in her left shoulder and arm. She had no motor weakness in the manual muscle testing of both the upper and lower extremities, and deep-tendon reflexes of the arms and legs were normal with no bilateral ankle clonus. Magnetic resonance imaging (MRI) showed an encapsulated intradural extramedullary mass at C4-C5. The mass was isosignal compared to spinal cord on T2-weighted imaging (T2WI) and was uniformly high signal on T1weighted imaging (T1WI) with the contrast medium (Figure 1). Preoperative differential diagnosis was considered the intradural extramedullary spinal cord tumor, neurinoma, neurofibroma, meningioma, metastatic tumor from ovarian cancer.

The patient underwent C4–C5 hemilaminectomy, tumor resection, and biopsy. The intradural extramedullary tumor had a thin capsule and was scarlet. Botryoidal vessels could be seen under the capsule, and there were no findings of tumor adhesion to the arachnoid and dura mater. The origin of the tumor seemed to be in the left C5 posterior rootlet. Histological examination of the resection specimen from the tumor revealed an adenocarcinoma with a neural tissue papillary formation, confirming the diagnosis of ovarian cancer metastasis (Figure 2).

After surgery, her intolerable neck-shoulder pain was fully resolved, and she had no difficulties with daily living activities. However, two months later she underwent gamma knife radiosurgery for a resection of a recurrent metastatic brain tumor, and four months later she died from cachexia.

Discussion

To our knowledge, this is the third report in the literature that describes a patient with intradural extramedullary spinal cord metastasis from ovarian cancer (Table 1).^{7,8} Intradural spinal cord metastasis is considered the result of "drop metastasis" from brain metastasis from lung, prostate, breast, and melanoma.^{5,9,10} On the other hand, some reports have been published on cases of metastatic intradural extramedullary spinal tumors with no brain metastasis.^{11–16} Our patient had a history of metastatic brain tumor from ovarian cancer. Regardless of having brain metastases, the possibility of metastatic intradural extramedullary spinal tumors should be considered in patients having carcinoma.

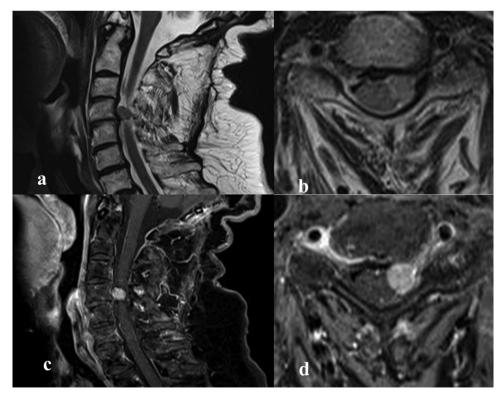


Figure 1 Sagittal T2WI (a) and axial (b), Sagittal T1WI with contrast medium (c) and axial (d) cervical spine MR images. The mass was in the C4-C5 intradural extramedullary space. The mass showed an iso-signal on T2WI (a, b) and homogeneous enhancement with contrast medium on T1WI (c, d).

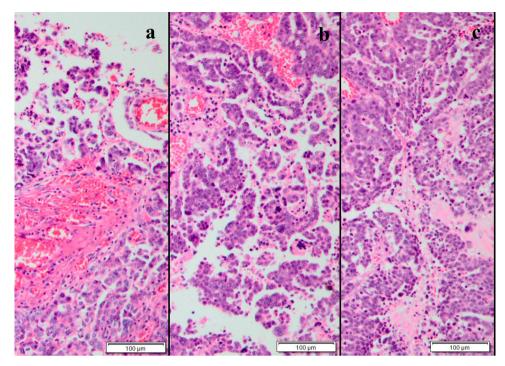


Figure 2 A hematoxylin and eosin-stained tissue section showing serous adenocarcinoma with papillary proliferation: (a) ovary; (b) brain; (c) cervical spinal rootlet.

There are two published concepts of intradural extramedullary metastases. The first is called "drop metastasis." Here, metastatic spread from intracranial lesions can disseminate neoplastic cells through the cerebrospinal fluid (CSF).⁶ In patients without intracranial lesions, there are five routes as the second concept: (1) hematogenous, via the arterial system, (2) via the perineural lymphatics, (3) through the venous plexus, (4) spreading via the subarachnoid space, and (5) seeding from involved osseous structures to the CSF through the dura mater¹⁷ Because our patient had a history of metastatic brain tumor, the most reasonable route was through CSF, the so-called "drop metastasis". However, some reports have reported on cases of metastatic intradural extramedullary spinal cord tumors without brain metastases. Therefore, four of the five routes, except for (5), mentioned above were also plausible in our case.

The most common primary intradural extramedullary spinal cord tumors are derived from sheath cells covering the spinal nerve roots (schwannomas and neurofibromas) or meningeal cells located along the spinal cord surface (meningiomas) (6). Schwannomas and neurofibromas are indistinguishable on MRI. Pre-contrast T1-weighted sequences failed to detect intradural extramedullary metastatic tumors. Hence, a post-contrast T1-weighted sequence is the optimal modality as a diagnostic tool for intradural extramedullary metastatic tumors (10). In our case, the C4–C5 intradural extramedullary mass showed homogeneous enhancement with contrast medium on T1-weighted images (T1WI). An MRI with a contrast medium on T1WI is adequate for the preoperative diagnosis of metastatic intradural extramedullary spinal tumor.

Because intradural extramedullary metastasis usually indicates an advanced widespread progression of systemic malignancy, aggressive surgical resection of lesions is usually not done. Frey (10) described that the duration between diagnosis and death in 30 patients with intradural extramedullary secondary neoplasms of less than one

Table 1 Metastatic intradural extramedullary spinal cord tumor from ovarian cancer: previous report summary.

Author, Year	Age (years)	Level	Enhancement on T1WI	Other metastasis	Postop outcome	Survival time
Chow et al.7	64	T7–T8	+	Unknown	Unchanged	22 days
Mehrotra <i>et al.</i> 8	46	T8–T10	Not done	Unknown	Improved	Unknown (>3 weeks)
Our case	76	C4–C5	+	Brain	Improved	4 months

T = thoracic. C = cervical. T1WI = T1-weighted magnetic resonance imaging. Postop outcome = measured the day after surgery. Survival time = duration from surgery to death.

month was 43% (13/30 patients) and less than three months was 73% (22/30 patients) resulting in few cases of intradural extramedullary metastatic tumors occurring, and such cases should be recognized as the terminal stage of the primary cancer. Metastatic spinal tumor in the intradural extramedullary from ovarian cancer has a prognosis of 22 days to 4 months, meaning that in this case the stage was terminal. Therefore, it is critically important to have an accurate preoperative diagnosis for decision-making regarding treatment.

In general, nonsurgical management of metastatic spine disease is recommended when tumor involvement has not resulted in spinal instability, neurologic deficits, or pain nonresponsive to medical management.¹⁸ Radiation therapy would be the most appropriate management given the indications above.¹⁹ In this case, the preoperative differential diagnoses were Schwannoma, neurofibroma, or metastatic tumor. The possibility of a metastatic tumor was all but unthinkable because there were only two reports of metastatic intradural extramedullary spinal cord tumor from ovarian cancer in the literature. The main purpose of this resection was to eliminate intractable pain. The second purpose was to diagnose enhancing, intradural, and extramedullary lesions. CSF cytology may show evidence of malignant cells.¹⁰ However, CSF cytology is not always positive in patients with intradural extramedullary spinal metastases.²⁰ CSF cytology was positive in only two of ten patients with intradural extramedullary spinal metastases.⁹ Therefore, if CSF cytology is negative, histological examination of the resection specimen might be needed to achieve a definitive diagnosis.

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Conflicts of interest None.

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