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Solitary Metastatic Cauda Equina Tumor From Breast Cancer

-Case Report-

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Abstract

A 45-year-old woman presented with a rare case of metastatic cauda equina tumor from breast cancer without spinal column or brain metastasis. She had undergone resection of breast cancer 4 years previously. She presented with a 2-month history of severe low back pain. Magnetic resonance imaging showed a well-enhanced intradural extramedullary mass at the L1 level without other intradural lesions. At surgery, the tumor was partially removed to preserve the nerve root function under electrophysiological monitoring. The histological diagnosis was adenocarcinoma. The tumor was located in the subarachnoid space, suggesting hematogenous metastasis from the breast cancer. Postoperatively the pain subsided and no neurological deficit occurred. She underwent adjuvant therapy and rehabilitation. Cauda equina tumors may be relatively progressive regardless of imaging findings and clinical symptoms, so preoperative systemic investigation should be conducted, considering the possibility of metastatic tumor. A comprehensive therapeutic strategy involving adjuvant therapy after surgery is important to establish, considering the preservation of postoperative nerve function.

Key words: breast cancer, cauda equina, metastatic tumor

Introduction

Most tumors of the cauda equina are benign and occur as a primary lesion. Among 31 patients with cauda equina

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tumors, histological findings suggested neurofibroma in 14, ependymoma in 11, hemangioblastoma in 2, astrocytoma in 2, lipoma in 1, and epidermoid tumor in 1.⁶) No metastatic tumor was found. Brain and bony metastasis including the spinal column frequently occurs in patients with malignant tumors outside the central nervous system.^{11,12} However, intradural spinal cord metastasis is relatively rare. Symptomatic metastatic intramedullary tumors involving the cervical and thoracic cords were detected in 0.3% of autopsied cancer cases.³ Generally, brain metastatic tumors disseminate to the vertebral body or to the spinal cord. We encountered a case of solitary metastasis from breast cancer to the cauda equina without metastasis to the brain or vertebral body.

Case Report

A 45-year-old woman had a 2-month history of low back pain and motor weakness of the right lower extremity. She had undergone resection of breast cancer with preservation of partial round resection and level-II axillary lymph node dissection 4 years ago. She had received hormonal therapy with anti-estrogen agents for 2 years. Thereafter, she discontinued consulting the physician based on selfassessment. After moving to a new address, she visited a local clinic, and underwent examination of tumor markers. Since the CA15-3 level had increased, plain chest radiography and computed tomography of the lungs were taken, which revealed bilateral masses suggesting metastasis. Neither mammography nor ultrasonography of the primary focus showed any recurrence. Then, low back pain occurred. She complained of radiating pain and weakness of the right lower extremity 2 months later. Lumbar magnetic resonance (MR) imaging revealed an intradural extramedullary mass in the cauda equina. She was referred to our department for further examination and treatment.

On admission, she complained of low back pain projecting to the right S1 distribution, which decreased at rest. She had motor weakness in the right lower extremity (2-3/5 in the manual muscle test) and reduced right Achilles tendon reflex. No vesicorectal disorders were identified. Straight leg raising test was limited on the right. Serological examinations showed increases in the carcinoembryonic antigen, CA125, and CA15-3 levels. Plain lumbar radiography revealed no abnormalities. Lumbar MR imaging demonstrated a well-enhanced intradural extramedullary mass measuring $15 \times 18 \times 30$ mm with a clear border at the L1 level. T_1 - and T_2 -weighted imaging detected hyperintensity at the bottom of the tumor, suggesting intratumoral hemorrhage (Fig. 1). No metastasis to the spinal column or the central nervous system of both the brain and spinal cord was detected.

L1 laminoplastic laminotomy with T12-L2 partial laminectomy was performed for histological diagnosis and tumor removal. A reddish tumor was found in the subarachnoid space. The tumor was adherent to the peripheral cauda equina and encased a S1 nerve root (Fig. 2). Intraoperative electrophysiological stimulation of the cauda equina indicated that this encased nerve root innervated the gastrocnemius and anal sphincter muscles. Latency of the bulbocavernosus reflex significantly prolonged during tumor removal. The highly vascular tumor was difficult to dissect from the surrounding neural structures. To preserve the vesicorectal function, the tumor was intentionally left along the encased nerve root. The intraoperative histological diagnosis was adenocarci-

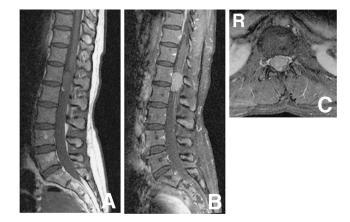


Fig. 1 A: Lumbosacral sagittal T_1 -weighted magnetic resonance image showing the intradural extramedullary mass as hyperintensity at the L1-2 level. B, C: Sagittal (B) and axial (C) T_1 -weighted magnetic resonance images with gadolinium showing a homogeneously enhanced lesion.



Fig. 2 Intraoperative photograph showing a reddish tumor in the subarachnoid space, adhered to the peripheral cauda equina and encasing a S1 nerve root.

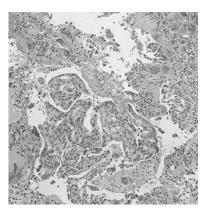


Fig. 3 Photomicrograph of the mass showing a tumor identical in appearance to adenocarcinoma. Hematoxylin and eosin stain, original magnification \times 200.

Author (Year)	Age (yrs)	Sex	Primary lesion	Location	Duration until operation (mos)	Lung lesion	Procedure	Sacrificed nerve	Neurological deterioration
Takahashi et al. (1990) ¹⁴⁾	51	М	RCC	L4	12	+	total	+	+
Maxwell et al. (1999) ⁹⁾	84	Μ	RCC	L2-3	2	+	total	+	—
Takada et al. (2003) ¹³⁾	61	Μ	RCC	L3	10	+	partial	+	+
Kubota et al. (2004) ⁸⁾	68	Μ	RCC	L3	15	+	total	+	_
Blondet et al. (2005) ²⁾	21	Μ	lung carcinoid	L3-4	6	+	total	NA	_
Kotil et al. (2007) ⁷⁾	50	Μ	lung adenoma	L4-5	0.5	+	total	+	_
Present case	45	F	breast cancer	L1	2	+	partial	-	_

Table 1 Summary of reported cases

NA: not available, RCC: renal cell carcinoma.

noma, possibly from the breast cancer (Fig. 3).

After surgery, the radiating pain of the right lower extremity and low back pain were relieved. The patient was referred to another hospital for treatment of the metastatic pulmonary lesions identified preoperatively and rehabilitation. The patient has undergone adjuvant chemotherapy and focal radiation therapy to the conus medullaris and pulmonary lesions without development of further neurological deficits.

Discussion

Table 1 summarizes the present case and the 6 previous cases of cauda equina metastasis in the absence of spinal column and brain metastasis.^{2,7-9,13,14} Primary lesions were renal cell carcinoma in 4 patients, lung cancer (adenocarcinoma, undifferentiated carcinoma) in 2, and breast cancer (adenocarcinoma) in 1. The present case was a metastasis from breast cancer. All 7 patients had primary pulmonary lesions or lung metastases from the primary focus.

Metastatic cauda equina tumors do not have typical initial symptoms, but are characterized by the rapid speed of the symptom progression (Table 1). The mean interval from the appearance of symptoms until surgery was 6.8 months (2 weeks to 15 months). In our patient, low back pain and radiating pain of the lower extremity had also worsened over 2 months. The mean interval between the appearance of symptoms and the diagnosis of primary benign cauda equina tumor was 39 months in 31 patients.⁶⁾ Review of 70 patients with primary cauda equina tumor found that the interval was 2 years or more in 41 patients, and 5 years or more in 21 patients.⁴⁾ A diagnosis of cauda equina tumor was made within 6 months in only 8 patients. The progression of metastatic cauda equina tumors is faster than that of primary benign cauda equina tumors, indicating high malignancy. The symptoms caused by cauda equina tumor occurred as initial symptoms in 2 patients, leading to the confirmation of the primary focus.7,14)

Preoperative MR imaging has no characteristic findings. Many studies indicated that the tumor has a clear border with homogeneous enhancement. However, MR imaging is not useful for differentiating metastatic cauda equina tumors from neurinoma or ependymoma, which are frequently found in the cauda equina. Furthermore, intratumoral hemorrhage, as observed in our patient, was confirmed in 10% of patients with neurinoma or ependymoma. The specificity, as a finding suggesting malignancy, may be low.¹¹

Five routes have been hypothesized for the dissemination of metastatic intradural spinal tumor from outside the central nervous system: (a) hematogenous, via the arterial system; (b) via the perineural lymphatics; (c) through the rich venous plexus; (d) spreading via the subarachnoid space; and (e) seeding from the involved osseous structure to the cerebrospinal fluid through the dura mater.^{5,8)} Metastatic intracranial tumors are concomitantly detected in 90% of patients with metastatic intradural spinal cord tumors.¹⁰⁾ In many cases, the metastatic intradural spinal cord tumors may be disseminated as drop metastases via the subarachnoid space. Systemic hematogenous metastasis has been excluded in patients with cauda equina metastasis without other spinal cord metastasis, and metastasis via the paravertebral venous plexus (Batson venous plexus) considered likely.9,13) The paravertebral venous plexus includes no venous valve, and communicates with the portal vein, inferior vena cava, and azygos vein.¹⁾ Therefore, an increase in the abdominal pressure might result in venous blood regurgitation, so tumor cells could migrate to the spinal cord from the pelvis, thoracic or abdominal organs without passing through the lung, forming a metastatic focus. On the other hand, epidural lesions were observed in most patients with lymphogenous metastasis or metastasis via the spinal venous system.¹⁴⁾ The lymphogenous and venous metastasis might not occur in patients with cauda equina metastasis without epidural lesion. Concurrent pulmonary lesions were common among the 7 patients including our patient with cauda equina metastasis. Neither concomitant epidural lesions nor other spinal metastatic lesions were detected. Therefore, cauda equina metastasis may have occurred via an arterial hematogenous route.

The nerve roots must be carefully managed during tumor removal. In contrast to primary tumors of the cauda equina, the nerve roots, which may retain intact function, are surrounded by the metastatic tumors. Excluding one patient with no reported intraoperative findings, the tumor involved some nerve roots in all patients (Table 1). Postoperative neurological condition deteriorated in 2 of the 5 patients in whom the tumor was removed by cutting the nerve roots during surgery.^{13,14)} Motor weakness of the lower extremity was noted in one patient, and sensory disturbance of the cut nerve area occurred after surgery in the other patient. Low back pain and radiating pain of the lower extremity subsided, and no neurological deficit developed postoperatively in the remaining 3 patients.

In our patient, intraoperative electrophysiological stimulation of the cauda equina showed contractions of the gastrocnemius and anal sphincter muscles. Latency of the bulbocavernosus reflex was prolonged during tumor removal. Therefore, the tumor was maximally removed with preservation of the nerve roots surrounded by the tumor. Postoperatively, low back pain subsided, and no additional neurological deficit occurred. Intradural metastatic cauda equina tumors may disseminate to other tissues of the central nervous system via the cerebrospinal fluid. Cure is difficult to achieve by only surgery, so postoperative adjuvant therapy should be performed in accordance with the histological type of the primary focus. Therefore, if metastatic tumors are suspected, even the lesion is malignant, cauda equina nerve function should be maintained with partial removal under intraoperative electrophysiological monitoring. Adjuvant therapy is needed after tumor removal.

Cauda equina tumors may be relatively progressive regardless of imaging findings and clinical symptoms, so preoperative systemic investigation should be conducted, considering the possibility of metastatic tumor. The primary focus must be investigated, and a histological diagnosis obtained. A comprehensive therapeutic strategy involving adjuvant therapy after surgery is important to establish, considering the preservation of postoperative nerve function.

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