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Case Report

Sacral Nerve Root Metastasis in a Patient with Lung Carcinoma Resembling Neurinoma – a Case Report and Literature Review

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Abstract

Intradural extramedullary metastases from systemic neoplasms are very rare, with an incidence ranging from 2% to 5% of all secondary spinal diseases. We present the case of a 53-year-old man diagnosed with lung adenocarcinoma with symptoms of severe back pain and tibial paresis. The magnetic resonance imaging (MRI) revealed an intradural lesion originating from the right S1 nerve root mimicking neurinoma. Total tumor removal was achieved via posterior midline approach. The histological examination was consistent with lung carcinoma metastasis. Due to the rarity of single nodular nerve root metastases, MRI images may be misinterpreted as nerve sheath tumors, such as schwannomas or neurofibromas. We performed a brief literature review outlining the mainstay of diagnosis, therapeutic approach, and the prognosis of these rare lesions.

Keywords

intradural, leptomeningeal metastasis, MRI, nerve root, spine, surgery

INTRODUCTION

With an incidence ranging from 2% to 5% of all secondary spinal illnesses, intradural extramedullary metastases (IEM) from systemic neoplasms are extremely uncommon. They can be either solitary or multiple.[1]

IEM are observed in cases of malignant cell dissemination into the cerebrospinal fluid through the leptomeninges. Although the evidence for IEMs is limited to a few case reports or small case series, they are considered to be predominantly epithelial in origin. The primary sources are most commonly breast carcinoma (12%–35%), lung carcinoma (10%–26%), and gastrointestinal tract carcinoma (4%–14%).[2]

In addition to providing a brief literature analysis of cases of nerve root metastasis that have been published, the current research aims to present the case of a patient with lung cancer who had both nodular nerve root metastasis and intracerebral and leptomeningeal spinal metastases (LSM).

CASE REPORT

We present the case of a 53-year-old male patient with histologically verified lung adenocarcinoma who underwent chemotherapy and radiation therapy. Six months later, he presented with onset of severe back pain that radiated toward the posterior surface of the right leg followed by acute weakness of plantar flexion of the right foot. The neurological examination on admission to hospital revealed severe back pain (VAS 10/10), radicular hyperalgesia (VAS 10/10), and

hyperesthesia along the right S1 dermatome. He also had tibial nerve plegia of the right foot, muscle weakness measured 0/5 with inability for plantar flexion. Noncontrast magnetic resonance imaging (MRI) revealed an intradural extramedullary tumor at the S1 level, originating from the nerve root, which was initially interpreted as neurinoma ***(Figs 1A-C)****.*

The patient was operated on via typical midline posterior surgical access. The right S1 nerve root was found to be thickened. Meticulous dissection between the root fibers was performed ***(Fig. 1D)****. The tumor was tightly adherent to one of the fibers* ***(Fig. 1E)****. By means of microsurgical technique, the tumor was completely removed* ***(Fig. 1F)****. The histological examination confirmed metastasis from undifferentiated small cell neuroendocrine lung carcinoma* ***(Fig. 2)****.*

During the postoperative period, the radiculopathy was partially resolved. The weakness of the right foot persisted but the pain was gradually reduced to 2/10 according to VAS. On the second postoperative day, the patient reported headache and loss of vision in both eyes and left eyelid drop. A new set of neurological symptoms that pointed to cerebral involvement prompted us to perform a contrast-enhanced MRI of the brain and spine axis. It revealed multiple intracerebral metastases,

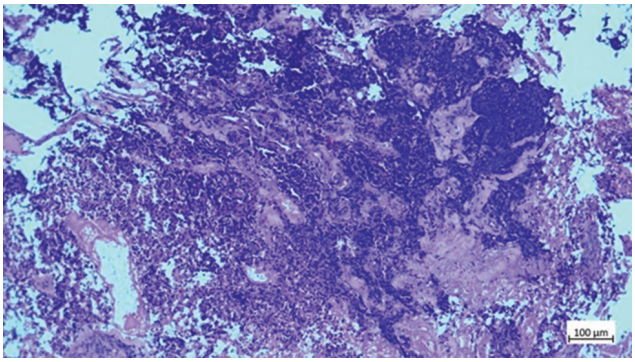

Figure 2. Histological examination verifying undifferentiated small cell neuroendocrine lung carcinoma (Hematoxylin-Eosin staining, ×100).

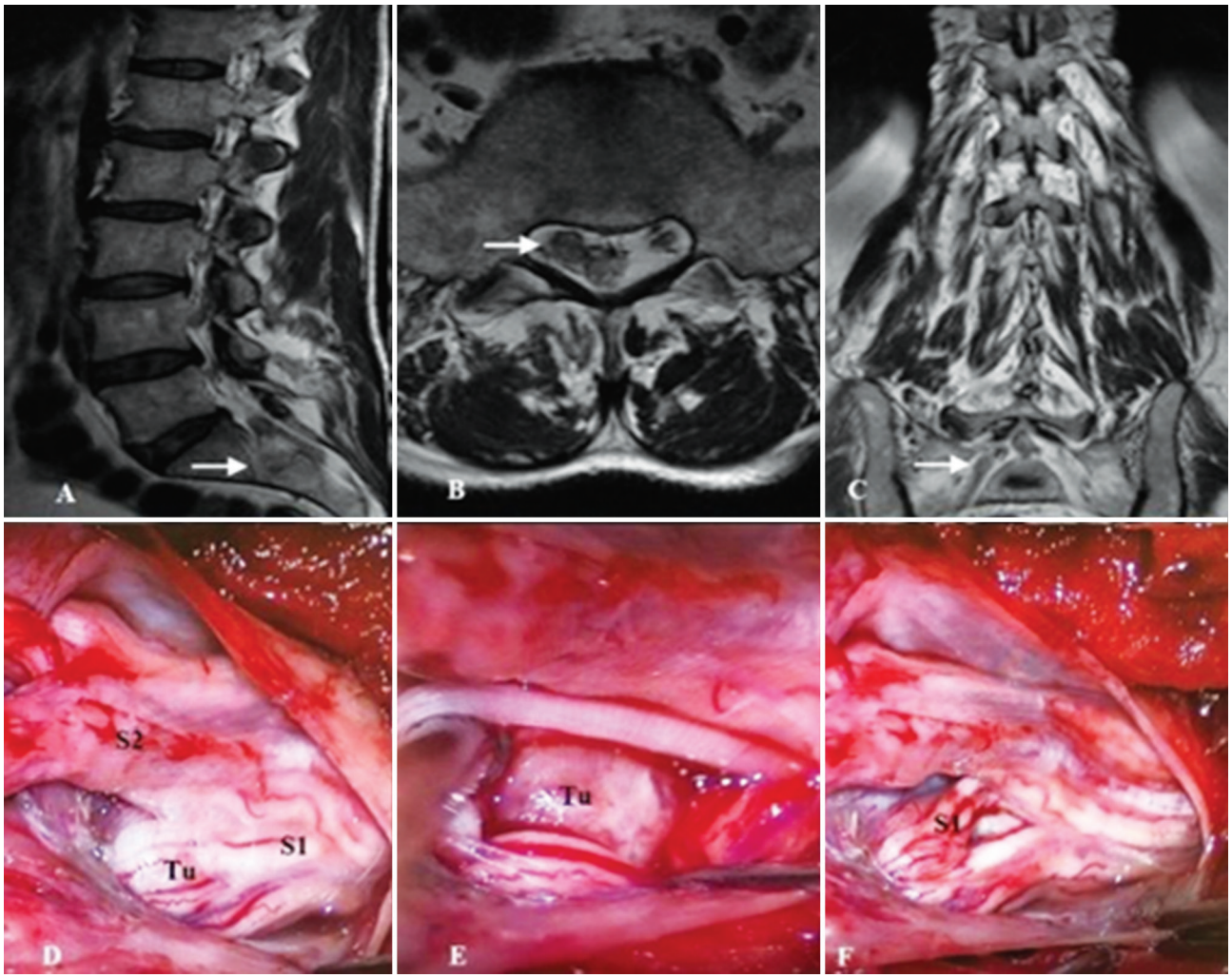

Figure 1. (A-C): Pre-operative T2-weighted MRI demonstrating an isointense intradural tumor mass in the right S1 nerve root on sagittal, axial, and coronal projections (arrows); (**D-F**): Intraoperative images illustrating important steps of the microsurgical tumor removal. Tu: tumor; S1: first sacral nerve root; S2: second sacral nerve root

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including several lesions located in the optic nerve and optic chiasm, as well as other multiple intradural "drop" metastases in the cervical and lumbar spinal segments ***(Figs 3, 4)*** *which were omitted in the initial preoperative non-contrasted MRI. The patient was followed up until the third postop month when he passed away.*

DISCUSSION

We present a concise review of nerve root metastases reported in the literature and initial clinical symptoms **(Table 1)**.

Lung carcinoma is the most common malignant disease, accounting for approximately 34% of all male and 22% of

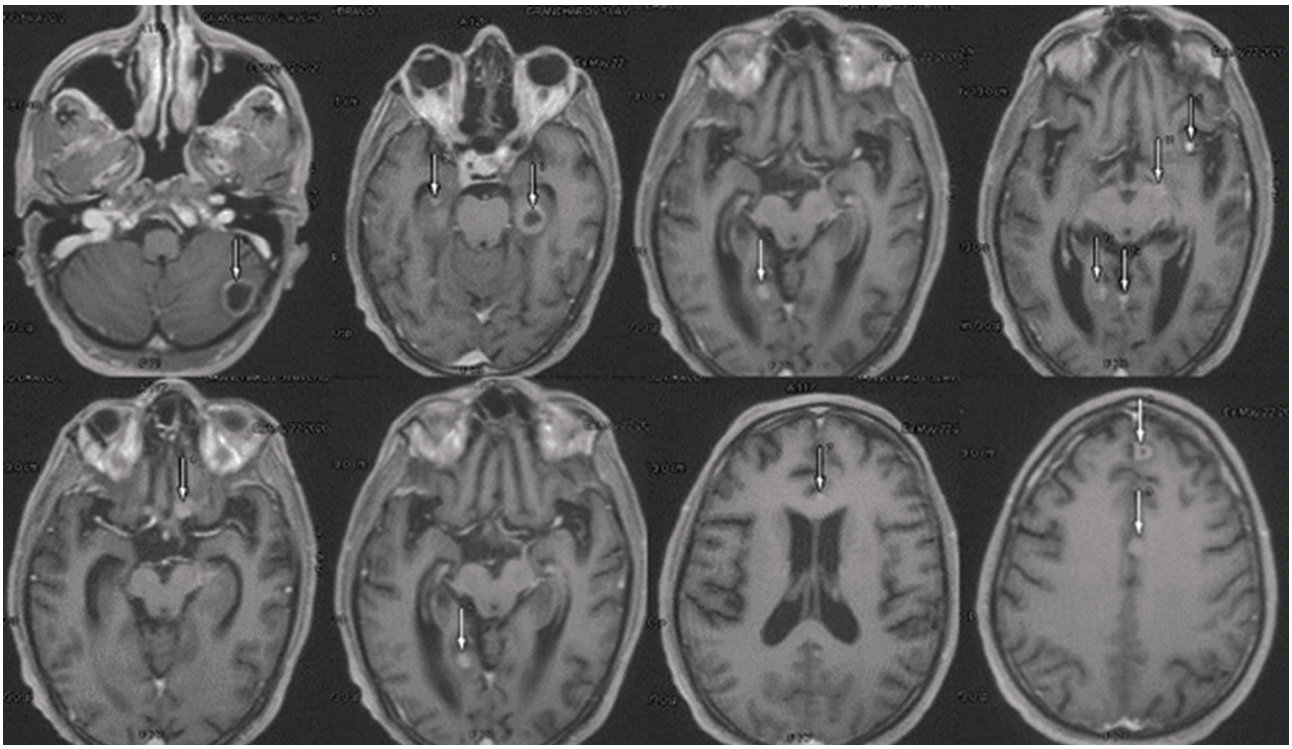

Figure 3. Contrast-enhanced brain MRI revealed multiple intracranial metastases (arrows).

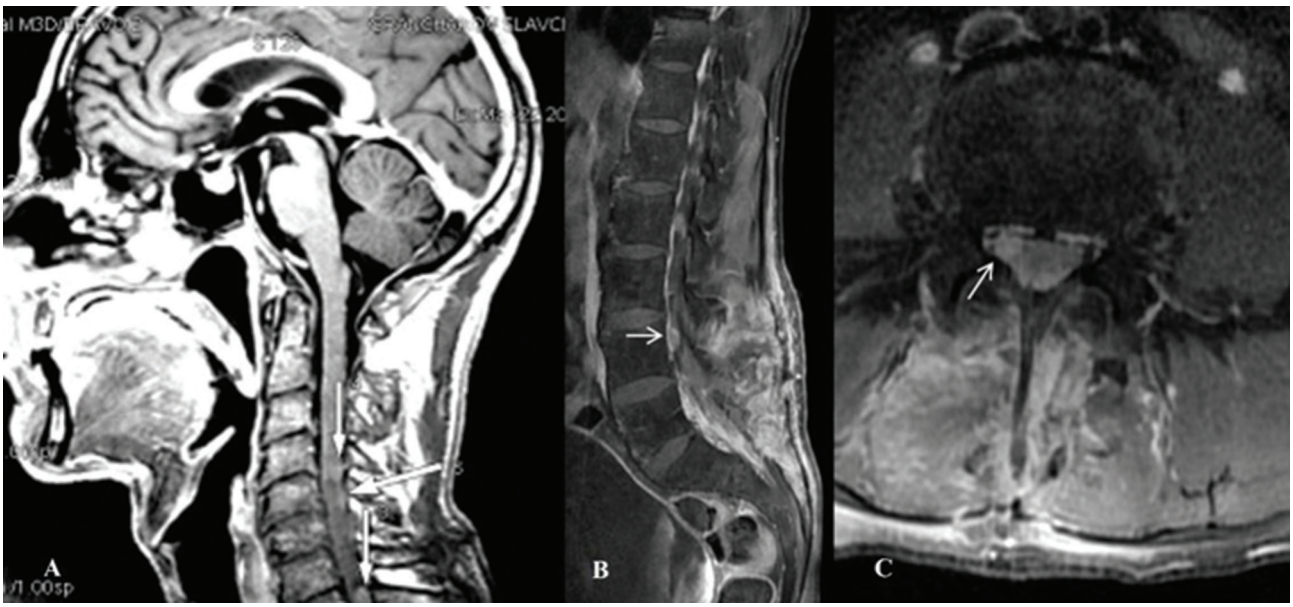

Figure 4. Postoperative contrast-enhanced brain and spine MRI: **A**. Sagittal T1-weighted cervical MRI demonstrating intradural extramedullary metastases at C4–C5, C8, and Th1 levels (arrows); **B**, **C**. Sagittal and axial T1-weighted lumbar MRI visualize intense contrast enhanced nodule at the level of L4 vertebrae (arrows).

Table 1. Brief overview of published cases with lumbo-sacral nerve root metastasis.[3-16]

Author	Primary Source	Affected nerve root	Symptoms
Johnson et al.[3]	Colonic adenocarcinoma	N/A	No symptoms
Johnson et al.[3]	Lung carcinoma	N/A	Foot drop
Wigfield et al.[4]	Colonic adenocarcinoma	L1	Pain syndrome
Uchida et al.[5]	Uterine adenocarcinoma	S1 and S2	Low back pain; radicular pain and numbness
Schulz et al.[6]	Breast carcinoma	L2	Radicular pain
Mitchell et al.[7]	Ewing sarcoma	L4	Low back pain; radicular pain and numbness
Ito et al.[8]	Breast carcinoma	S1	Low back pain; leg muscle weakness
Cabrilo et al.[9]	Renal cell carcinoma	L5	Low back pain; radicular pain and numbness
Slotty et al.[10]	Lung adenocarcinoma	L3	Radicular pain and numbness
Strong et al.[11]	Renal cell carcinoma	L4	Radicular pain and numbness
Li et al.[12]	Squamous cell carcinoma	L5/S1	Radicular pain
Oktay et al.[13]	Lung adenocarcinoma	S1	Low back pain; leg muscle weakness, numbness, and hyperalgesia
Di Sibio et al.[14]	Gastric adenocarcinoma	S1	Low back pain; radicular pain and numbness
Zhang et al.[15]	Breast carcinoma	S1	Radicular pain
Zhang et al.[15]	Breast carcinoma	L5 and bilateral cervical roots	Radicular pain and numbness
Norouzi et al.[16]	Breast carcinoma	S1	Radicular pain
Our case	Lung neuroendocrine carcinoma	S1	Low back pain; radicular pain; acute tibial nerve plegia

all female cancer deaths and most commonly metastasizes in the central nervous system (20-25%), the cervical lymph nodes (15-60%), bones (25%), and the liver (10-15%).[13] The incidence of metastatic lesions in the peripheral nervous system is very low. According to Jaeckle, the lumbosacral plexus is affected in 0.71% and the brachial plexus - in 0.43% of cases.[17] Nerve root metastases are extremely rare and are commonly falsely interpreted as nerve sheath tumors, such as schwannomas or neurofibromas, as was in our case.[14]

The possible routes for cancer cell dissemination to the nerve roots are arterial, venous or lymphatic spread, local invasion through the dura, and dissemination via the cerebrospinal fluid (CSF) into the subarachnoid space, so-called "drop metastases" or LSM.[18] Multiple LSM usually spread through the CSF, especially in the presence of intracranial metastases, similar to the presented case.[10]

The clinical presentation of LSM includes cauda equina syndrome, communicating hydrocephalus, and cranial neuropathies. Early in the course of the disease, neurological symptoms can be minimal, such as radiculopathy or visual disturbances. As mentioned, we also observed damage to second and third cranial nerves in addition to S1 nerve root.[19]

There are two types of LSM distribution – diffuse and nodular. The diffuse type is characterized with free floating non-adherent cancer cells, while the nodular consists of leptomeningeal tumors that tend to enhance with gadolinium on MRI.[20] According to Chamberlain et al., in 30% to 70% of cases, non-contrast MRI fails to diagnose LSM.[19] Contrast-enhanced MRI is the gold standard for diagnosing LSM.[21] According to Palmisciano et al., most cases of LSM are associated with intracranial metastases.[21] As demonstrated in our case, a contrast-enhanced MRI of the brain and spinal cord should be performed for establishing the number and location of lesions if neurological symptoms of LSM occur.

While diffuse LSM can remain asymptomatic, nodular spinal metastases usually cause nerve root compression, resulting in radicular pain, radiculopathy, or cauda equina syndrome.[21,22] In the presented case, both types of LSM were diagnosed.

LSM can occur at all spinal levels. According to Carminucci and Hanft, and Mariniello et al., LSM are rare in the area of the cauda equina, while Palmisciano et al. report that in this area they are most common, which is explained by the slow circulation of cerebrospinal fluid.[21,23,24] The presence of LSM is associated with advanced stage of the neoplastic disease and indicates poor prognosis.[24] According to Palmisciano et al., the median overall survival is approximately 3 months, which is confirmed by our case.[21] The median time between primary cancer diagnosis and metastatic dissemination is 12 months.[21]

The presented case is an example of the development of early metastatic lesions (in less than 1 year) after applying multidisciplinary treatment with radiation and chemotherapy. This fact could be explained by the loss of receptors, susceptible to chemotherapeutic drugs, and progression of the most sustainable and undifferentiated tumor cells.[25]

In cases of solitary tumors with slow growth, LSM can be diagnosed prior to the primary neoplasm.[26]

Most authors recommend surgical decompression and partial or total metastasis removal in patients with acute motor deficit or cauda equina syndrome.[13,18,24,27] The goal of surgery is to relieve the neurological symptoms and thus improve the quality of life.[21]

The presence of multiple intracerebral metastases and LSM requires an assessment of the risk of possible postoperative complications. We hypothesize that in our case, the ocular symptoms that developed on the second postoperative day may have resulted from the excessive CSF drainage during the intradural part of the surgery, which led to acute compression of the optic and oculomotor nerves by cerebral metastasis.

CONCLUSION

LSM are late complications of systemic cancer that significantly worsen the quality of life of patients and are associated with high mortality. When an intradural extramedullary tumor is detected in the region of the cauda equina in a patient with previously diagnosed systemic cancer, possible metastasis should be included in the differential diagnosis. In these cases, a contrast-enhanced MRI of the brain and spine axis should be performed to establish or exclude the presence of parenchymal and/or LSM. The risk of complications following palliative surgical intervention should be carefully assessed for each individual case.

Author contribution

Conception or design of the study: B.K.; data collection: P.A.; data analysis and interpretation: I.K.; drafting the manuscript: B.K.; critical revision of the manuscript: I.K.; other (study supervision, fundings, materials, etc.): P.A.; final approval of the version to be published: I.K.

Patient informed consent

Informed consent was obtained from all individual participants included in this study.

Conflict of Interest

There is no conflict of interest to disclose.

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Метастазы в корень крестцового нерва у пациента с карциномой лёгкого, напоминающей невриному: описание случая и обзор литературы

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Резюме

Интрадуральные экстрамедуллярные метастазы системных новообразований встречаются очень редко, их частота составляет от 2% до 5% всех вторичных заболеваний позвоночника. Мы представляем случай 53-летнего мужчины с диагнозом аденокарцинома лёгкого с симптомами сильной боли в спине и пареза большеберцовой кости. Магнитно-резонансная томография (МРТ) выявила интрадуральное поражение правого нервного корешка S1, имитирующее невриному. Полное удаление опухоли было достигнуто задним срединным доступом. Гистологическое исследование подтвердило метастазы рака лёгкого. Из-за редкости единичных узловых метастазов в нервных корешках изображения МРТ могут быть ошибочно приняты за опухоли оболочек нервов, такие как шванномы или нейрофибромы. Мы провели краткий обзор литературы, в котором изложены основы диагностики, терапевтического подхода и прогноза этих редких поражений.

Ключевые слова

интрадурально, лептоменингеальные метастазы, МРТ, нервный корешок, позвоночник, хирурги