

## Case Report

# Cauda equina syndrome caused by isolated spinal extramedullary-intradural cauda equina metastasis is the primary symptom of small cell lung cancer: a case report and review of the literature

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**Abstract:** Objectives: To describe a case of extramedullary-intradural metastases causing cauda equina syndrome (CES) as the primary syndrome of lung cancer, and to review the pertinent medical literature. Summary of Background Data: Intradural spinal metastasis is rare, accounting for 6% of all spinal metastases. Of all primary lesion types, lung carcinoma is about 40-85%. Extramedullary-intradural metastases of lung cancer causing CES is unusual. Methods: A 55-year-old patient with symptoms of low back pain that radiated to bilateral lower legs with painful paresthesias and piecemeal sphincter disturbances and lead to urinary incontinence, and constipation at last for three months. The MRI showed that there was an isolated, well-demarcated, intradural extramedullary mass at the L3, 4 levels. Results: The patient was admitted to the hospital as the severe cauda equina syndrome (CES) and considering the possibility of nerve system neoplasms for surgery. After a series of examinations, the X-ray and computed tomography (CT) detected a tumor in the left upper lobe with hilar and mediastinal lymph node metastases. The concluded diagnosis through histopathologic examination with immunohistochemistry after the patient received an L3-L4 laminectomy and tumor excision and biopsy was extramedullary-intradural spinal metastasis of the small cell lung cancer (SCLC). The patient had a rapid improvement of the CES. Conclusion: Although the majority of cauda equina tumors are primary tumors, extramedullary-intradural metastasis should be considered before surgery in patients with rapid developed CES.

**Keywords:** Cauda equina syndrome, small cell lung cancer, extramedullary intradural spinal metastasis

## Introduction

Metastasis at intradural-extramedullary cauda equine caused by lung cancer was very rare [1]. Once the neoplasm metastasize to the cauda equina, symptoms such as low back pain, bilateral sciatica, motor weakness of the lower extremities, and rectal and bladder sphincter dysfunction may occur and is therefore clinically diagnosed as cauda equina syndrome (CES) [1-3]. To our knowledge, the isolated metastasis cases at intradural-extramedullary cauda equine caused by SCLC were not found before the detection and therapy of SCLC [3]. This report describes the case of a 55-year-old man who developed rapid cauda equina syndrome then was confirmed as the small cell lung cancer by radiologic check and histopathologic examination with immunohistochemistry.

## Case report

A 55-year-old man with symptoms and signs of low back pain that radiated to bilateral lower legs with painful paresthesias and piecemeal sphincter disturbances for three months was admitted to our hospital in September 2011. The patient had a smoking history of about 30 years and had quit smoking for about 5 years. The patient presented with low back pain when fasciitis was initially considered. Conservative therapy such as rest and massage is preformed. They could work a little. Eight weeks later, painful paresthesias and dysesthesias spread to the posterior area of thighs, buttocks and the perianal area with progressively worsened conditions. In clinics, the patient's lumbosacral magnetic resonance imaging (MRI) revealed an extramedullary intradural mass measuring 2.6

## CES is the primary symptom of small cell lung cancer



**Figure 1.** The lumbosacral magnetic resonance imaging showing an extramedullary-intradural mass (white arrows) measuring 2.6 cm craniocaudally by 1.2 cm coronally (A) and 1.1 cm anteroposteriorly in horizontal plane (B) at the L3-L4 level. The mass demonstrated relatively low signal intensity on T2-weighted-image in the sagittal (A) and axial (B) plane.



**Figure 2.** The contrast enhanced lumbosacral magnetic resonance imaging showing an extramedullary intradural mass (white arrow) demonstrated the mild homogeneous enhancement, a clear boundary and relatively intermediate signal intensity on T1-weighted image in the sagittal plane.

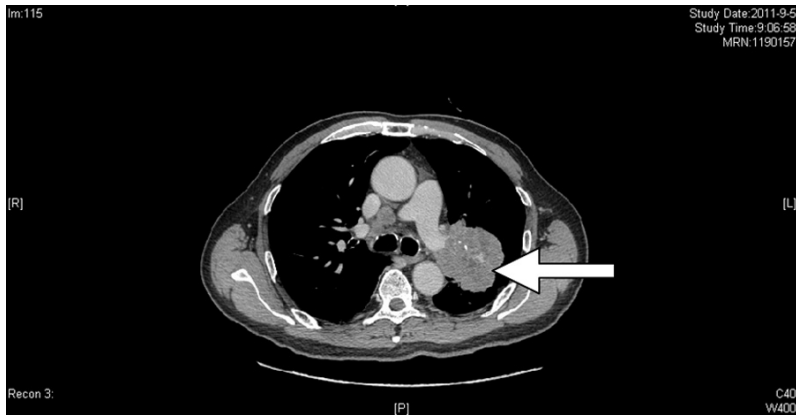
cm craniocaudally, 1.2 cm coronally by 1.1 cm anteroposteriorly in the horizontal plane at the L3, 4 level (**Figure 1A** and **1B**). He was admit-

ted to the hospital considering the possibility of nerve system neoplasms for surgery in September 2011. During this time, the constipation sustained in conjunction with urinary incontinence. The contrast enhanced MRI showed the mild homogeneous enhancement and a clear boundary for the mass (**Figure 2**). The neurologic examination showed no decreased muscle tone and strength in the legs. Hamstring and quadriceps abductor and adductor strength of his legs were 4/5. Ankle flexor and extensor strengths were measured to be 5 bilateral. Bilateral Babinski signs were not elicited. Bilateral deep tendon reflexes decreased. Sensation of vibration, joint position, pinprick, and temperature was impaired in both legs. Bilateral elevation test of straight leg were positive at 45°. There were tenderness percussion pain at spinous processes of L3-5. Radiating pain was obvious while doing that. The thoracic cord, cervical cord and brain MRI scans showed no evidence of tumor.

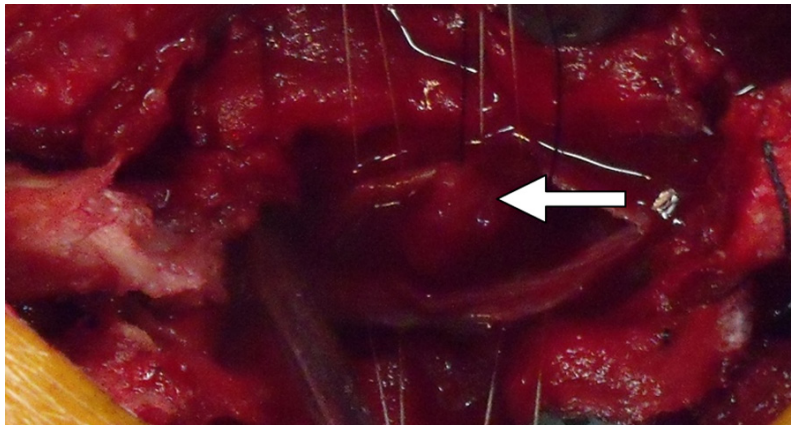
The preoperative routine chest radiograph showed an abnormal mass in the left lung. A chest computed tomography (CT) scan revealed a tumor in the left upper lobe with ipsilateral hilar and mediastinal lymph node metastases (**Figure 3**). There is no evidence of other metastasis according

to bone scan. The patient was radiographically and clinically diagnosed as lung cancer (clinical stage IV) by the supporting findings.

## CES is the primary symptom of small cell lung cancer



**Figure 3.** A chest computed tomography (CT) scan revealed a mass (white arrows) in the left upper lobe with ipsilateral hilar and mediastinal lymph node metastases.



**Figure 4.** The picture showed a pale pink fragile mass, which infiltrated and surrounded the majority of the caudal rootlets. There is no obvious erosion to nerve.

The patient underwent an L3-L4 laminectomy and internal fixation because of the serious pain and CES (constipation and urinary incontinence) firstly. After opening dural, we found a pale pink fragile mass, which infiltrated and surrounded the majority of the caudal rootlets. There is no obvious erosion to nerve (**Figure 4**), so we resected the tumor as most as we could. The pathologic section of extramedullary-intradural spinal metastasis showed the tumor cells arranged like nest and flakes and were beam shaped and adenoid structured. There were hemorrhage and necrosis partly. It's obvious that there were small round and oval cells with nuclear molding. The cells were characterized by hyperchromatic nuclei with scanty cytoplasm, ill-defined cell borders. The SCLC was diagnosed by histopathologic examination with

HE and immunohistochemistry staining (CK7(partly+), CK20(-), TTF-1(+++), SyN(+), CgA(partly+), Vimentin(-), CK(focus+), EMA(partly+), S-100(-), GFAP(-), Ki-67(70%+)) (**Figure 5A-C**).

Two weeks after the surgery, treatments of combination chemotherapy with cisplatin and etoposide were performed. The wound healed and there was no pain any more. He could urinate himself after removing the catheter. After the first course of chemotherapy, the patient was discharged from our hospital in September 2011. And the patient was followed up regularly and died after one year.

### Discussion

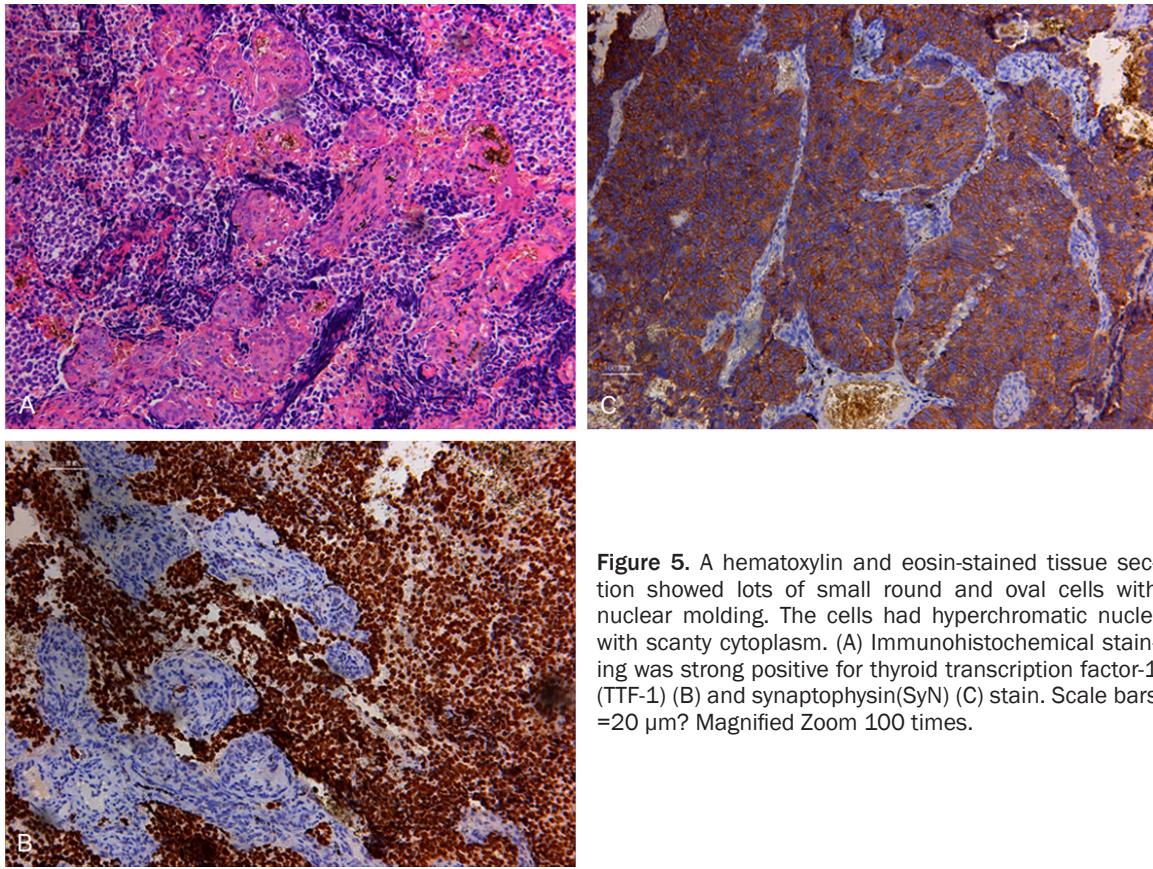
CES is the clinical diagnosis of polyradicular symptomatology caused by the compression of the lumbosacral nerve roots located in the dural sac [1-3]. Spinal neoplasms were the main causes of the CES. Different spinal neoplasm causing the CES have different loca-

tions, such as epidural, intramedullary, and extramedullary intradural [4-6]. Most of patients were affected by epidural spinal cord compression. The majority of isolated cauda equina tumors causing CES are primary neurologic tumors such as glioma and meningioma [7]. Extramedullary-intradural cauda equina metastatic tumors from outside the central nervous system are very rare [1-8]. Up to now, there are 8 cases, including our current case, being reported as the cauda equine metastases caused by lung cancer. (see **Table 1**) 20 cases, including this case, have been reported in the literature. Renal cell carcinoma was the most common source of cauda equine metastasis in all the reported cases [9-32].

All patients had the diagnosis with previous cancer history to differentiate spinal cauda



## CES is the primary symptom of small cell lung cancer



**Figure 5.** A hematoxylin and eosin-stained tissue section showed lots of small round and oval cells with nuclear molding. The cells had hyperchromatic nuclei with scanty cytoplasm. (A) Immunohistochemical staining was strong positive for thyroid transcription factor-1 (TTF-1) (B) and synaptophysin(SyN) (C) stain. Scale bars =20  $\mu$ m? Magnified Zoom 100 times.

equina metastasis from primary neoplastic lesions. Our reported case here has no cancer history until the detection of CES. CES is the primary syndrome. He also has no proof of other metastasis. In this case, we should make the diagnosis with clinical history, MRI and the exploration of surgery. The syndrome had a quick clinical progression within 3 months. MRI showed that there was a isolated mass in cauda equine with clear edge. However, because of the rapid progress in CES, a diagnostic surgery of decompression and biopsy of the mass lesion was preformed firstly. This released the CES. At the same time, we got the clear diagnosis of the mass. Our case is a rare case with metastasis at intradural-extramedullary spinal cord caused by small cell lung cancer. SCLC is very aggressive in clinical progression, with the inclination of frequent widespread metastases. But to the best of our knowledge, this is the first reported case of isolated extramedullary intradural spinal metastasis of SCLC causing CES which was the the primary syndrome of lung cancer.

A decompression surgery mostly of laminectomy is necessary when a patient develops cauda

equine syndrome with the injury of sphincter function. As what was said in the literature (**Table 1**), most of the surgeries could give the patients a obvious improve in the syndromes even if possibly it couldn't help to improve the survival time and avoid the complications of spinal fluid leak, hematoma, postoperative infections, failure to resect the tumor completely et al. [4]. Specially to SCLC patient with extramedullary intradural spinal metastasis, the tumor is aggressive. The immediate surgery is still recommended following the rapid developed CES [4, 5].

Up to now, there were 5 possible routes for metastatic intradural spinal tumor from outside the central nervous system [1-9]. They are the rich venous plexus route; perineural lymphatics route; seeding route from involved osseous structures to the cerebrospinal fluid through the dura; spreading route via subarachnoid space; and hematogenous via the arterial system route [3, 9-32]. We tried to find the possible route through which our reported case metastasized. Because all the auxiliary examination revealed no evidence of bony, abdominal and cerebral metastasis in our case, hema-

## CES is the primary symptom of small cell lung cancer

**Table 1.** [3] Patient profiles in some reported cases of intradural extramedullary cauda equine spinal metastasis of lung cancer

Author	Age/sex	level	interval	Num (Sol./Mul.)	Clinical syndrome	Brain Invol.	Pathology of lung cancer	Therapy	Outcome	Fellow-up time
Coutinho LM [1]	60/M	Cauda equina	3 ms	Sol	pain	unknown	undifferentiated carcinoma	Surg	unknown	unknown
Lin CL [3]	79/M	L2-3	1 m	Sol	CES+pain+	no	SCLC	Surg	Death	1 m
Kotil K [10]	50/M	L4/5	2 ws	Sol	CES	no	adenocarcinoma	Surg	Resolved	1 ys
ERIC BLONDET [11]	23/M	L3-4	6 ms	Sol	Pain	no	atypical lung carcinoid	Surg	Improved	2 ys death
Cansever T [12]	79/M	L2-3	1y	Sol	CES+pain	no	pulmonary squamous cell	Surg	Improved	unknown
Chow TS [13]	61/M	L3-4	Average 3.6 ms	Unknown	sensory changes	unknown	adenocarcinoma	Surg	Improved	6 ms death
Tsimpas A [14]	62/M	L2	4 ms	Sol	Pain+weakness	unknown	Large cell neuroendocrine carcinoma	Surg	Pain release	12 ms
Present case	55/M	L3-4	3 ms	Sol	CES+Pain	no	SCLC	Surg	Death	12 ms

togenous metastasis seem to be the most possible way to spread. Small cell lung cancer is predisposed to hematogenous spreading. All of the reported cases had had metastasis or treatments of lung cancer before the unusual metastasis to the cauda equine. But our case is a case of extramedullary intradural metastases causing cauda equine syndrome (CES) as the primary symptom of lung cancer. Maybe that means this small cell lung cancer is more aggressive. We must watch out the unusual metastasis to the cauda equina in lung cancer cases especially in SCLC cases. At the same time, we should find the possible evidence of metastasis in extramedullary-intradural tumor cases when we make the diagnosis. The extramedullary-intradural metastases causing cauda equine syndrome (CES) maybe is the sign of deterioration of SCLC.

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## Disclosure of conflict of interest

None.

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## CES is the primary symptom of small cell lung cancer

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