# **CASE REPORT**

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# Cervical intramedullary spinal cord metastasis from esophageal adenocarcinoma: a case report

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# Abstract

**Background** Intramedullary spinal cord metastasis (ISCM) is a rare pathologic entity. To the best of our knowledge, this is the second case of ISCM due to esophageal adenocarcinoma reported in the literature.

**Case description** We report a 63-year-old man with a history of surgical removal of an esophageal adenocarcinoma who presented with the Brown–Sequard syndrome. Pan-spinal MRI showed intramedullary cervical lesions with intense homogeneous enhancement on gadolinium injection at C3 level. There were other sites of spinal metastases and one in the brain. We proceeded with the surgical removal of the lumbar metastatic lesion. The patient died 10 days later from respiratory failure.

**Conclusion** ISCM from esophageal adenocarcinoma appeared as homogeneous high intensity of the intramedullary lesion in T1 gadolinium. Their prognosis is very poor.

Keywords Intramedullary spinal cord, Metastasis, Cervical, Esophageal adenocarcinoma, Case report

## Introduction

The occurrence of intramedullary spinal metastases (ISCM) from esophageal cancer is extremely rare. The most frequently reported subtype of primary esophageal tumor is squamous cell carcinoma (SCC) [1]. In 2015, Dalkilic et al. reported a case of ISCM from esophageal adenocarcinoma [2]. However, ISCM's overall prognosis is very poor. Here we describe a 63-year-old man with Brown–Sequard syndrome by ISCM of an esophageal adenocarcinoma with detailed clinical and radiological data.

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# Case report

# **Patient information**

A 63-year-old man was admitted on February 21, 2023, with neck pain and progressive upper and lower extremity weakness on the right side of the body that has been developing for four months. In November 2021, the patient underwent transhiatal surgical resection of a moderately differentiated esophageal adenocarcinoma of the lower third of the esophagus. Postoperatively, he underwent cycles of chemotherapy with 5-fluoroura-cil (5-FU) and cisplatin prior to admission. He had not received radiation therapy.

# **Clinical findings**

On neurological examination, we found Brown–Sequard syndrome. The patient had a normal muscle tone with a Medical Research Council (MRC) scale for muscle strength of 1/5 in both upper and lower right limbs, and



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proprioceptive deficits on the right side of the body and loss of pain and temperature sensation on the left side of the body. His bilateral patellar and ankle clonus and Babinski's sign were positive. He had a bladder dysfunction. The paravertebral muscles of the cervical spine were contracted. There was no spinal deformity. In addition, the patient's general condition was impaired, and the Karnofsky Performance Status (KPS) was estimated at 50%. The diagnosis of cervical spinal cord compression was suspected.

## **Diagnostic assessment**

Magnetic resonance imaging (MRI) of the whole spine was performed. Sagittal STIR revealed a highintensity intramedullary lesion extending from C2 to C4 (Fig. 1A), and intense homogeneous enhancement ovoid images in T1 with gadolinium at C3 level (Fig. 1C). At the dorsal and lumbar levels, imaging showed diffuse spinal leptomeningeal lesions suggestive metastasis (Fig. 2A, B). Cerebral MRI showed left temporal metastatic lesion in axial T1 Flair after



Fig. 1 A Sagittal STIR magnetic resonance imaging showed a high-intensity area spanning from C2 to C4 (yellow star). B Axial T2-weighted showed a high-intensity intramedullary lesion on the right side of cervical hemicord (yellow arrow). C High intense ovoid images enhanced in T1 Gado at C3 level (yellow arrow)



Fig. 2 Sagittal T1-weighted post-gadolinium enhanced imaging showed intense lesions suggestive metastasis at dorsal (A) and lumbar (B) level (yellow arrow). Cerebral MRI showed left temporal metastatic lesion (C) in axial T1 Flair after gadolinium injection (yellow arrow)

gadolinium injection (Fig. 2C). Additional biological tests were performed, but without any particularity. The carcinoembryonic antigen (CEA) level was 6.83 ng/ml (normal range < 5 ng/ml).

## Therapeutic intervention

We discussed in a multidisciplinary consultative meeting between neurosurgeons, oncologists and anesthesiologists to proceed with surgical removal of the lumbar metastasis lesion for pathological analysis, improving distal neurologic function, and ultimately the quality of life. The patient underwent a laminectomy of L1 and L2, and a durotomy at the corresponding level. We found an adherent grayish intradural lesion, extramedullary, easily resectable. The duroplasty was watertight. Pathologic analysis was suggestive of a metastasis of a moderately differentiated adenocarcinoma with CK7- and CK20positive cytokeratin immunohistochemical (IHC) profiles (Fig. 3), indicating an upper gastrointestinal tract origin.

## Follow-up and outcomes

In the immediate postoperative course, the patient maintained the same motor and sensory deficits. The radiotherapy session was scheduled after surgical wound healing. Seven days after the operation, the patient showed worsening of breathing. He was transferred to the department of the intensive care unit. He died 3 days after being admitted to the intensive care unit.

## Discussion

ISCM from esophageal cancer is extremely rare. Only five cases of ISCM due to esophageal cancer have been reported in the literature [1]. The most common subtype of primary esophageal cancer is squamous cell carcinoma (SCC) [1]. To our knowledge, this is the second report of intradural spinal metastases from esophageal adenocarcinoma.

The cervical localization of ISCM is the most common localization in the literature [1, 4]. This could be related to its larger mass and richer vascular supply [5]. In our case, ISCM also occurred in the cervical cord. Three hypotheses have been put forward to explain the pathophysiology of ISCM: hematogenous spread, meningeal carcinomatosis and direct invasion [3]. The most accepted is hematogenous spread, which may explain why metastasis can occur at any level of the spinal cord. This theory seems conclusive since our patient was also battling and multifocal spinal metastases.

The clinical symptoms are similar to all spinal cord compressions that result in a lesion. It depends on the degree of injury. The most frequently found signs are back or neck pain, weakness of the extremities, loss of sensitivity, urinary incontinence [4]. In our case, the patient had Brown–Sequard syndrome. This neurological syndrome results from hemisection of the spinal cord. The diagnosis of ISCMs mainly depends on the patient's medical history and imaging findings [5]. MRI is considered the most sensitive method for diagnosing ISCM. There are no specific imaging criteria to distinguish intramedullary



Fig. 3 A The analyzed fragments microscopically showed cribriform tumor proliferation. The cells have a hyperchromatic and nucleolus. B The atypia are moderates. By IHC the tumor cells are positive for CK7 and CK20, suggesting metastasis of a moderately differentiated adenocarcinoma

metastases from different types of spinal tumors. Some authors reported two enhancement features on MR imaging for non-central nervous system intramedullary spinal cord metastases: a more intense thin rim of peripheral enhancement around an enhancing lesion (rim sign) and an ill-defined flame-shaped region of enhancement at the superior/inferior margins (flame sign) [6, 7]. Shen et al. reported mixed intense tumor with typical, more intense, thin border of peripheral enhancement. These are similar characteristics for the ISCM secondary to SCC [5]. However, our case showed that this observation cannot be generalized. The lesion had a homogeneous intensity after gadolinium injection. Under these conditions, we can extrapolate that the radiological features of ISCM due to esophageal adenocarcinoma are high-intensity and homogeneous lesions after being enhanced by gadolinium injection.

There are no established guidelines for treating ISCMs. Radiation therapy is the gold standard of therapy for ISCM [1, 3, 5]. In selected cases, an operation can be performed. We opted for a surgical approach in the aim of confirming the diagnosis of metastasis, improving distal neurologic function, and ultimately the quality of life. The choice of the lumbar region for surgery of the metastasis was guided by the concern to avoid the risk of secondary worsening, including the life-threatening risk in the upper cervical spinal cord. ISCM is a known harbinger of end-stage cancer, and patients with multiple lesions that includes the brain tend to have poor surgical outcomes [8]. ISCM's overall prognosis is very poor; the mortality rate is 80% in the 3 to 4 months after the onset of the first symptoms [5]. KPS is a reliable parameter used in primary tumor pathologies and in metastases to predict survival. One study showed that low KPS (<50%) was strongly associated with death within a relatively short period of time [9, 10]. Our patient's case did not escape the rule.

# Conclusions

ISCM from esophageal cancer is extremely rare. We described the second case of cervical ISCM due to esophageal adenocarcinoma revealed by Brown–Sequard syndrome. MRI remains the gold standard of diagnosis although there are no specific radiological characteristics of intramedullary metastases. Nevertheless, our study reported a homogeneous high intensity of the intramed-ullary lesion in T1 gadolinium. Because of its rarity, there is no guideline for treating ISCM due to esophageal cancer. Their prognosis is worse.

#### Abbreviations

ISCM Intramedullary spinal cord metastases SCC Spinal cord compression 5-FU 5-Fluorouracil MRC3 Medical Research Council MRI Magnetic resonance imaging STIR Short Tau inversion recovery CEA Carcinoembryonic antigen IHC Immunohistochemical CK Cytokeratin

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Nil

## Author contributions

FLOO did conceptualization, writing draft, reviewing and editing, visualization, supervision, validation, methodology. IEK done writing, review and editing. YCHD was involved in writing, review and editing, JL contributed to writing, review and editing. MES, ACEA and MS done writing and editing. MG performed supervision, validation, and review. All authors read and approved the final manuscript.

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#### Availability of data and materials

All data generated or analyzed during this study are included in this published article.

## Declarations

#### Ethics approval and consent to participate

The informed consent of the patient's family was obtained prior to submission of this article. In addition, this article follows both the Consensus-based Clinical Case Reporting Guideline and the Recommendations for the Conducting, Reporting, Editing, and Publication of Scholarly Work in Medical Journals.

## **Consent for publication**

The patient's family has given informed consent to the publication of their case.

### **Competing interests**

The authors declare that they have no competing interests.

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