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## Case Report

# A rare presentation of leiomyosarcoma metastasis to the cervical spine: A case report and a brief review

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## ABSTRACT

**Background:** Leiomyosarcoma (LMS) is a rare malignancy that originates from smooth muscle. The most common sites of metastases include the lungs, liver, kidney, and skin. Notably, metastases of LMS to the central nervous system/or spine are extremely rare. When a cervical spinal LMS lesion was encountered, the patient successfully underwent gross total tumor resection with negative margins.

**Case Description:** A 63-year-old female had undergone an anterior cervical C5–C7 discectomy and fusion 18 years ago and resection of a retroperitoneal LMS 3 years ago. She newly presented with right-sided numbness and pain of 2 months duration that correlated with a focal right-sided C5-level hemiparesis (i.e., 4/5 motor strength). When the cervical magnetic resonance demonstrated a right-sided C5 intralaminar mass with extension into the C5–C6 foramen, she underwent posterior tumor resection; pathologically, this proved to be an LMS metastasis. Respectively, 1- and six months postoperatively, follow-up magnetic resonance imaging scans showed no tumor recurrence; she tolerated adjuvant oncological treatment accompanied by physical therapy. However, in one postoperative year, the lesion recurred, and she is presently under consideration for additional surgical management.

**Conclusion:** Gross total surgical resection is the first line of treatment for patients with metastatic LMS. Here, a patient with a C5 lamina/C5–C6 foraminal bony LMS metastasis underwent posterior tumor resection accompanied by adjuvant oncological treatment but exhibited disease recurrence within one postoperative year.

**Keywords:** Leiomyosarcoma, Metastasis, Radiculopathy, Spine

## INTRODUCTION

Leiomyosarcoma (LMS) is a rare malignant tumor that arises from smooth muscle cells. It most commonly involves the uterus, followed by the gastrointestinal tract and retroperitoneum; its incidence is 0.7 cases per 100,000 patients/year.<sup>[1]</sup> Metastases of LMS typically spread to the lungs, liver, kidneys, and skin; it is only rarely encountered in the central nervous system or spine.<sup>[2]</sup> Metastatic LMS spinal lesions classically have a poor prognosis due to the high risk for tumor recurrence and relative resistance to radiation and/or chemotherapy.<sup>[8]</sup> Here, a 63-year-

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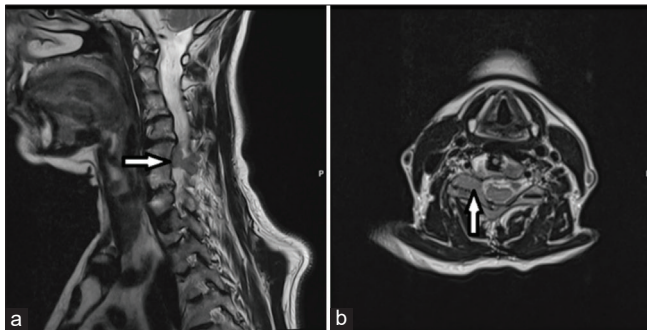
old female who, in 2021, had a retroperitoneal LMS resected newly presented with a right C5 level hemiparesis attributed to a C5 laminar/C5–C6 bony foraminal lesion. Following resection, the lesion proved to be a LMS metastasis that recurred within one year following gross total tumor removal despite adjuvant oncologic treatment.

## CASE REPORT

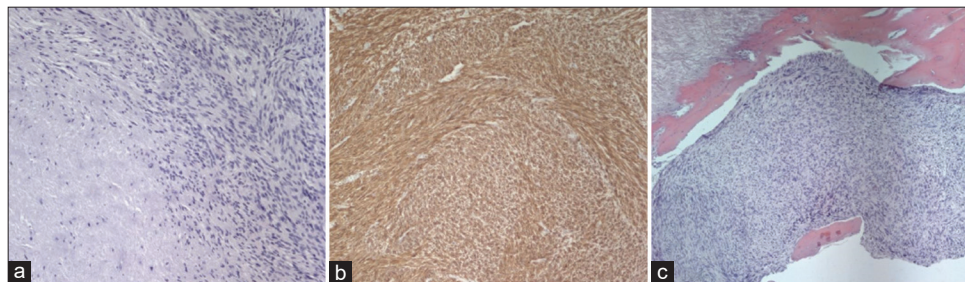
A 63-year-old female presented with a right mild hemiparesis (i.e., 4/5 motor deficit) and relative right-sided C5 sensory level of 1 month's duration. In 2005, she had a C5–C7 anterior cervical discectomy and fusion, while in 2021, she had a retroperitoneal LMS completely removed. She underwent posterior surgical resection of the right-sided C5 laminar/C5–C6 bony foraminal lesion documented by magnetic resonance imaging (MRI) that proved to be a LMS metastasis [Figure 1]. Immunostained slides and histological findings revealed atypical, polymorphic spindle cells with numerous mitoses and smaller foci of necrosis; cells were immunohistochemically positive for smooth muscle actin, muscle-specific actin, and desmin; marker CD34 was negative [Figure 2].

### Postoperative course

Postoperative cervical MRI scans revealed no tumor recurrence 1 and 6 months postoperatively [Figures 3-5].



**Figure 1:** Preoperative sagittal (a) and axial (b) T2-weighted MRI scans revealed the right-sided tumor mass lesion at the levels of C5 and C6. Arrows point to the primary tumor site on both images.



**Figure 2:** (a)  $\times 10$  magnification – Hematoxylin and eosin staining: Spindle-shaped cells with moderate nuclear pleomorphism, with area of necrosis, (b)  $\times 10$  magnification – Immunohistochemical staining, diffuse smooth muscle actin, positivity in tumor cells, and (c)  $\times 5$  magnification – hematoxylin and eosin staining, tumor infiltration, and destruction of the bone.

Postoperative follow-up computed tomography scan of the thorax, abdomen, and pelvis two months after surgery also revealed no additional metastatic tumors; the patient underwent adjuvant oncologic treatment (i.e., docetaxel and gemcitabine) during a timespan of 4 weeks. However, one year later, the C5 lesion recurred, and she is presently undergoing consideration for additional surgical treatment.

## DISCUSSION

### LMS rarely metastasizes to the spine

LMSs are highly malignant tumors that rarely metastasize to the spine. Sun *et al.*, in 2013, reported a metastatic LMS lesion to the cervical spine with destruction of the C6 vertebra; the patient underwent an anterior decompression and fusion.<sup>[8]</sup> In 2022, LiBrizzi *et al.* retrospectively analyzed bone metastases in LMS patients; most involved the femur, humerus, and spine; the latter were typically managed with kyphoplasty, decompression alone, and/or decompression and fusion.<sup>[4]</sup>

### Survival rates for spinal LMS

LMS are aggressive, highly malignant lesions that frequently recur and metastasize.<sup>[5]</sup> Elhammady *et al.*,<sup>[2]</sup> in 2007 and Ziewacz *et al.*,<sup>[10]</sup> in 2012 both presented large groups of patients with LMS; the five patients in the former study demonstrated a better overall survival rate of 8.6 years versus 10.7 month-survival for the nine patients in the latter series.<sup>[5]</sup>

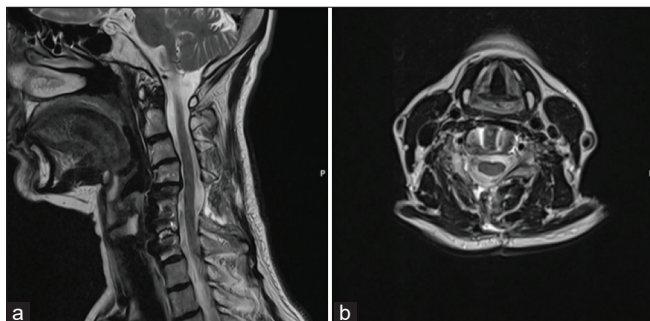
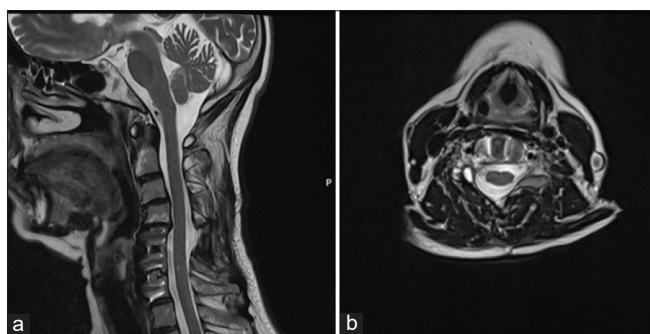
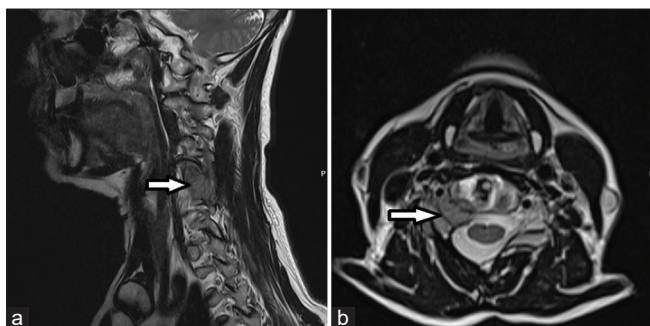
### Bony metastases for LMS lesions

LMS rarely causes osseous spinal vertebral metastases.<sup>[6]</sup> In their meta-analysis, Imura *et al.* concluded that chemotherapy could prolong survival in patients with LMS metastases who were ineligible for resection of metastatic lesions.<sup>[3]</sup> Recently, published studies documented several primary cervical spinal LMS lesions [Table 1] following both surgery and oncologic treatment in which the longest recurrence-free survival was 38 months.<sup>[7,9,10]</sup>

**Table 1:** Comparison of two recently published studies of primary LMS of the cervical spine with two metastatic cases.

Author/year	Radiologic findings/study	Treatment	Follow-up/outcome
Sun <i>et al.</i> , 2022	C3–C5, case report, primary LMS	GTR+ Radiotherapy	1 month, alive
Zhang <i>et al.</i> , 2022	C2–C3, primary LMS C4, primary LMS C2, primary LMS	Subtotal resection GTR GTR+Radiotherapy	6 months, progression-free survival 38 months, recurrence-free
Sun <i>et al.</i> , 2014	C6, metastasis	GTR+Radiotherapy	13 months, progression-free survival
Our study, 2023	C5, metastasis	GTR+Chemotherapy	12 months, recurrence-free survival

GTR: Gross total removal, LMS: Leiomyosarcoma

**Figure 3:** Postoperative sagittal (a) and axial (b) MRI T2-weighted scans one month after surgery revealed a complete tumor removal.**Figure 4:** Postoperative sagittal (a) and axial (b) MRI T2-weighted scans six months after surgery revealed a complete tumor removal, no tumor remnants and recurrences.**Figure 5:** Postoperative sagittal (a) and axial (b) MRI T2-weighted scans a year after surgery revealed tumor recurrence at the previous surgical site. Arrows depict sites of tumor recurrence on both images.

## CONCLUSION

The prognosis for patients with metastatic LMS is poor due to the high rate of metastases. Here, a 63-year-old female with a 2021 diagnosis of a retroperitoneal LMS was newly presented with a C5 laminar/C5–C6 foraminal bony lesion that, despite gross total posterior excision, recurred one year later.

## Ethical approval

The Institutional Review Board approval is not required.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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## Conflicts of interest

There are no conflicts of interest.

## Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript, and no images were manipulated using AI.

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