



Case Report

Delayed cervical spine metastasis from intracranial solitary fibrous tumor

Mohammad Mohsin Arshad¹, Arshad Ali¹, Abdalnasser Thabet¹, Issam A. AI-Bozom²

¹Department of Neurosurgery, Neuroscience Institute, ²Department of Pathology, Hamad Medical Corporation, Doha, Qatar.

ABSTRACT

Cervical spine metastasis from primary intracranial solitary fibrous tumors (SFTs) is an extremely rare clinical entity. This report focuses on its metastatic tendency, radiological imaging, management plan, and follow-up strategies in view of its long latency period for metastasis. A 35-year-old female presented with right-side cervical radiculopathy. Magnetic resonance imaging spine showed C7 vertebral body collapse with retropulsion and neural compression. Two years ago, the patient had surgical resection of intracranial SFT (World Health Organization grade 3) with no evidence of recurrence on follow-up imaging. Cervical C7 metastasis has been decompressed and fused by the anterior cervical approach. Histopathology confirmed SFT metastasis to the spine, and the patient received adjuvant radiotherapy. Cervical metastasis from well-controlled primary intracranial SFT poses a significant challenge for its diagnostic and management planning. Serial pre-emptive surveillance is warranted with regular imaging and appropriate patient counseling.

Keywords: Solitary fibrous tumor, Cervical, Anaplastic hemangiopericytoma, Metastasis, Intracranial, Spine, Delayed

INTRODUCTION

A solitary fibrous tumor (SFT) is a hypervascular sarcomatous tumor arising most likely from Zimmermann pericytes around the osseous capillaries and venules. Central nervous system (CNS) origin accounts for <1% of all CNS tumors and 2.4% of all meningeal tumors.^[2] Intracranial SFTs involving the spine are rare with only a few reports of isolated cervical spine metastasis.^[1,8,10,14-16]

We report a case of intracranial SFT with isolated cervical spinal metastasis after a 2-year dormant period. There was no evidence of metastatic lesions elsewhere including no local intracranial recurrence despite regular clinical and radiological screening. The scarcity of literature makes it difficult for a consensus management and follow-up plan. This report emphasizes metastatic propensities, radiological features, and management strategies, underscoring long-term clinical surveillance.

CASE REPORT

A 35-year-old female presented with neck pain, radiating to the right shoulder with weakness of the upper limb for the

last 10 days. Two years ago, she had surgical resection of the right temporal mass and the histopathology revealed SFT grade III (World Health Organization [WHO] classification 2021).^[7] Postoperatively, the patient received adjuvant radiotherapy (RT) and no recurrence on magnetic resonance imaging (MRI) brain after 2 years. On clinical examination, her distal muscle group had decreased power (grade 3/5) with impaired sensation in the right C7 and C8 dermatomes and exaggerated deep tendon reflexes. MRI cervical spine revealed decreased C7 vertebral height with mild retropulsion, with contrast enhancement, involving the right pedicle and encroaching bilateral exiting neural foramina, impinging on the nerve roots [Figure 1]. The positron emission tomography (PET) scan at 1 year was unremarkable for any metastatic disease.

The patient had a C7 corpectomy, with fusion using an expandable cage (ADDplus™, Ulrich Medical, Germany) and fixation by plating. The mass lesion was soft, grayish, and highly vascular. Histopathology revealed SFT grade 3 characterized by short fascicles of a monotonous population of ovoid cells with monomorphic nuclei, admixed with the background of variably sized and branched vasculature

*Corresponding author: Mohammad Mohsin Arshad, Department of Neurosurgery, Neuroscience Institute, Hamad Medical Corporation, Doha, Qatar. mohammadmohsinarshad@gmail.com

Received: 10 May 2023 Accepted: 25 June 2023 Epub Ahead of Print: 05 August 2023 Published: 10 November 2023 DOI: 10.25259/JNRP_252_2023

This is an open-access article distributed under the terms of the Creative Commons Attribution-Non Commercial-Share Alike 4.0 License, which allows others to remix, transform, and build upon the work non-commercially, as long as the author is credited and the new creations are licensed under the identical terms. ©2023 Published by Scientific Scholar on behalf of Journal of Neurosciences in Rural Practice

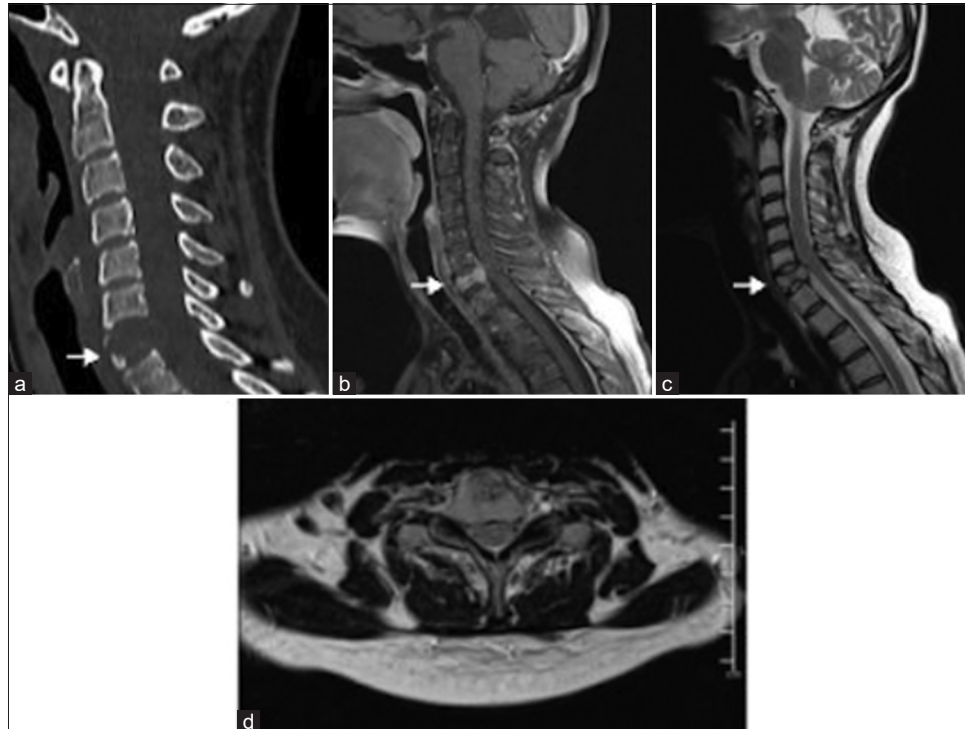


Figure 1: (a) Computed tomography sagittal view of the cervical spine showing a collapsed C7 vertebral body with retropulsion and expansile C7 vertebral body lesion, replacing most of the vertebral body. (b) T1 post-contrast and (c) T2-weighted sagittal and (d) Axial magnetic resonance imaging. Decreased vertebral height of C7 vertebra with mild retropulsion and intense post-gadolinium enhancement of the vertebral body and right pedicle.

called staghorn appearance [Figure 2]. Postoperatively, the patient got immediate pain relief and received adjuvant RT of the spine along with neurorehabilitation.

DISCUSSION

In 1949, Stout and Murray^[13] first described hemangiopericytoma (now called SFTs in the WHO classification 2021) as a rare vascular neoplasm. SFTs are aggressive tumors that tend to recur locally or distantly as extraneural distant metastases.^[6] CNS SFTs are uncommon and their extraneural distant spread to the spine is rarely reported.^[6,11] In 1961, Kruse^[5] reported intracranial SFTs that metastasized to the spine. Spinal metastasis usually occurs by one of three routes: Via lymphatic, direct extension, and hematogenous pathways.^[6] Valve-less connections between the intracranial venous system and the paravertebral venous plexus are the main pathways for spinal metastases.^[6,13] Differential spinal SFT diagnoses include malignant schwannoma, neuroblastoma, neurofibroma, and meningioma.^[11] In our case, although there was no local intracranial recurrence, and her surveillance PET scan was negative, isolated spinal metastasis still occurred after a 2-year latency period.

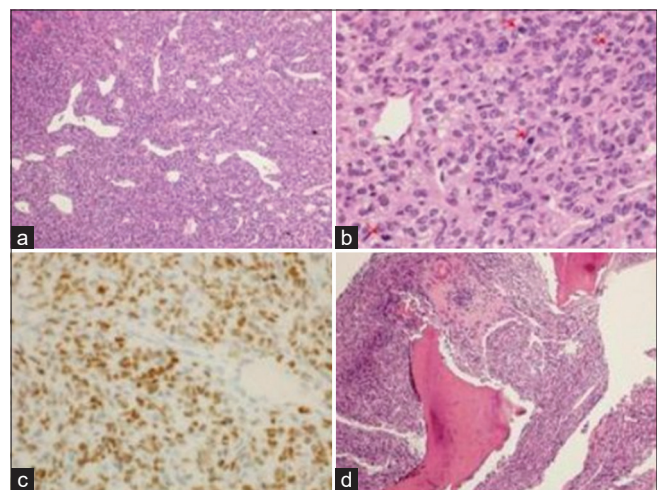


Figure 2: (a) Light microscopic examination of the tumor showing clusters of tumor cells in a background of staghorn blood vessels (H&E×100). (b) High-power view of the tumor showing monomorphic ovoid nuclei with numerous mitotic figures (red arrows) (H&E×400). (c) Immunohistochemistry with antibody against STAT6 diffusely expressed in the nuclei (immunohistochemistry×400). (d) Light microscopic examination of the recurrent tumor in the cervical spine showing bony trabeculae (red arrows) surrounded by tumor cells (H&E×100).

A PET scan is a useful tool to detect osseous metastasis at an early stage of disease progression, but a single negative scan may not be enough.^[6] The usual appearance of SFTs spinal metastasis is isointense to bone marrow on T1- and T2-weighted MR images, with homogeneous contrast enhancement, and the epicenter of the disease is usually in the vertebral body.^[9,11] Typical angiographic findings are a hypervascular mass with irregular, “corkscrew” vessels with a prolonged tumor blush and slow circulation time.^[12] In our case, an MRI scan showed a retropulsed C7 vertebra, intense post-gadolinium enhancement with an extension to posterior elements.

The management of spinal metastatic SFTs remains controversial, and surgical resection is the mainstay of treatment. There is no consensus on the extent of resection due to osseous invasion, and the proximity of vital neural tissues, especially in the cervical spine metastases.^[8,11] Pre-operative embolization has been used to reduce the risk of significant bleeding and facilitate maximal resection.^[12] Gamma Knife and CyberKnife have promising results as they increase survival and delay local recurrence.^[2] Gross total resection followed by adjuvant RT seems to be the best contemporary treatment strategy.^[2,4] High-precision RTs, such as fractionated stereotactic RT and intensity-modulated RT, are effective and safe.^[2] Radiosurgery and intensity-modulated RT can be considered in the setting of prior radiation, but chemotherapy remains controversial.^[4,11] The prognosis is poor once distal metastases have occurred and spinal metastases are, in particular, a rapidly progressive disease with acute neurological deficits.^[9] Combs *et al.*^[2] analyzed the use of high-precision RT (combined with surgical resection) in SFTs, including 2 spine cases, and demonstrated a survival rate of 100% at 5 years and 64% at 10 years.

It has been postulated that once the local cranial control of SFT is mitigated meticulously, spinal metastasis is likely to manifest due to a higher propensity for hematogenous spread via its peculiar valve-less angio-architecture.^[1,5,6,8] Extranural metastasis can occur with a different latency period which can be as much as several years, even though the primary lesion has been well controlled.^[5] In our case, this latent period was 2 years, but it has been reported as long as 11 years.^[14] Spinal metastasis should be kept in mind when a patient with adequately treated intracranial SFT presents with back or neck pain and/or is accompanied by limb weakness. Unfortunately, once symptomatic spinal metastases are diagnosed, there is no satisfactory treatment modality to stop their progression, and the prognosis remains poor.^[3,14] Therefore, it is essentially necessary to alert patients about possible spinal metastasis and the need for an extended clinical follow-up. The optimal strategy for surveillance is controversial, and a standard interval for serial imaging has not been established.^[4,6,10,13]

CONCLUSION

Intracranial SFTs are uncommon tumors with only a few reports of cervical spine involvement. Once a local cranial disease is well controlled by a diligent treatment strategy, there is a likely possibility of acutely progressive spinal metastatic disease even after a long latency period. This underscores the need for a pre-emptive strategy for serial imaging of the spine at regular intervals with appropriate counseling of patients to be vigilant for subtle clinical manifestations to ensure an early prompt diagnosis and management.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

- Cheon HC, Kim IY, Lee JK, Kim SH. Hemangiopericytoma with metastasis to the cervical spine: Case report. *J Korean Neurosurg Soc* 2003;33:102-4.
- Combs SE, Thilmann C, Debus J, Schulz-Ertner D. Precision radiotherapy for hemangiopericytomas of the central nervous system. *Cancer* 2005;104:2457-65.
- Dufour H, Métellus P, Fuentes S, Murracchiole X, Régis J, Figarella-Branger D, *et al.* Meningeal hemangiopericytoma: A retrospective study of 21 patients with special review of postoperative external radiotherapy. *Neurosurgery* 2001;48:756-63, discussion 762-3.
- Fountas KN, Kapsalaki E, Kassam M, Feltes CH, Dimopoulos VG, Robinson JS, *et al.* Management of intracranial meningeal hemangiopericytomas: Outcome and experience. *Neurosurg Rev* 2006;29:145-53.
- Kruse F Jr. Hemangiopericytoma of the meninges (angioblastic meningioma of Cushing and Eisenhardt). *Clinico-pathologic aspects and follow-up studies in 8 cases. Neurology* 1961;11:771-7.
- Lee JK, Kim SH, Joo SP, Kim TS, Jung S, Kim JH, *et al.* Spinal metastasis from cranial meningeal hemangiopericytomas. *Acta Neurochir (Wien)* 2006;148:787-90.
- Louis DN, Perry A, Wesseling P, Brat DJ, Cree IA, Figarella-Branger D, *et al.* The 2021 WHO classification of tumors of the central nervous system: A summary. *Neuro Oncol* 2021;23:1231-51.
- Nakashima H, Imagama S, Sakai Y, Nakamura H, Katayama Y, Ito Z, *et al.* Dumbbell-type hemangiopericytoma in the cervical spine: A case report and review. *J Orthop Sci* 2013;18:849-55.
- Okubo T, Nagoshi N, Tsuji O, Tachibana A, Kono H, Suzuki S, *et al.* Imaging characteristics and surgical outcomes in patients

- with intraspinal solitary fibrous tumor/hemangiopericytoma: A retrospective cohort study. *Global Spine J* 2023;13:276-83.
10. Ramdasi RV, Nadkarni TD, Goel NA. Hemangiopericytoma of the cervical spine. *J Craniovertebr Junction Spine* 2014;5:95-8.
 11. Rutkowski MJ, Jian BJ, Bloch O, Chen C, Sughrue ME, Tihan T, *et al.* Intracranial hemangiopericytoma: Clinical experience and treatment considerations in a modern series of 40 adult patients. *Cancer* 2012;118:1628-36.
 12. Santillan A, Zink W, Lavi E, Boockvar J, Gobin YP, Patsalides A. Endovascular embolization of cervical hemangiopericytoma with Onyx-18: Case report and review of the literature. *J Neurointerv Surg* 2011;3:304-7.
 13. Stout AP, Murray MR. Hemangiopericytoma: A vascular tumor featuring Zimmermann's pericytes. *Ann Surg* 1942;116:26-33.
 14. Woitzik J, Sommer C, Krauss JK. Delayed manifestation of spinal metastasis: A special feature of hemangiopericytoma. *Clin Neurol Neurosurg* 2003;105:159-66.
 15. Yi X, Xiao D, He Y, Yin H, Gong G, Long X, *et al.* Spinal solitary fibrous tumor/hemangiopericytoma: A clinicopathologic and radiologic analysis of eleven cases. *World Neurosurg* 2017;104:318-29.
 16. Zhang P, Jingmei HU, Dongsheng ZH. Hemangiopericytoma of the cervicothoracic spine: A case report and literature review. *Turk Neurosurg* 2014;24:948-53.

How to cite this article: Arshad M, Ali A, Thabet A, Al-Bozom IA. Delayed cervical spine metastasis from intracranial solitary fibrous tumor. *J Neurosci Rural Pract* 2023;14:750-3.