



Case Report

Atypical meningioma originating from the spinal accessory nerve

Hideaki Ueno¹, Satoshi Tsutsumi¹, Akane Hashizume², Natsuki Sugiyama¹, Hisato Ishii¹

Departments of ¹Neurological Surgery and ²Pathology, Juntendo University Urayasu Hospital, Urayasu, Japan.

E-mail: Hideaki Ueno - hideakiueno1229@gmail.com; *Satoshi Tsutsumi - shotaro@juntendo-urayasu.jp; Akane Hashizume - akane@juntendo-urayasu.jp; Natsuki Sugiyama - natsuking0602@yahoo.co.jp; Hisato Ishii - hisato-i@juntendo.ac.jp



*Corresponding author:

Satoshi Tsutsumi,
Department of Neurological
Surgery, Juntendo University
Urayasu Hospital, Urayasu,
Japan.

shotaro@juntendo-urayasu.jp

Received : 03 December 2022

Accepted : 20 December 2022

Published : 31 December 2022

DOI

10.25259/SNI_1085_2022

Quick Response Code:



ABSTRACT

Background: Atypical meningiomas rarely originate from the spinal accessory nerve at the C1–C2 level.

Case Description: A 57-year-old female presented with a 1-month history of headache. The cervical MR revealed a well-demarcated intradural/extramedullary tumor compressing the spinal cord at the C1–C2 level that measured 12 mm × 10 mm × 25 mm. She underwent microsurgical tumor resection. Intraoperatively, the tumor was adherent to the spinal accessory nerve, rather than the dura mater. Gross total tumor resection was performed, and the pathology was consistent with an atypical meningioma.

Conclusion: Atypical meningiomas rarely originate from the spinal accessory nerve. Gross total resection is the procedures of choice to mitigate the risk of tumor recurrence.

Keywords: Atypical meningioma, High cervical, Spinal accessory nerve, Spinal

INTRODUCTION

Spinal meningioma without apparent dural attachment has rarely been reported [Table 1].^[3-5,7] Here, a 57-year-old female with atypical meningioma at the C1–C2 level is presented that originated from the spinal accessory nerve.

CASE PRESENTATION

A 57-year-old female, without a history of neurofibromatosis, presented with a 1-month history of headache. The cervical magnetic resonance imaging (MR) revealed a right-sided, oval, and intradural/extramedullary lesion (i.e., 12 mm × 10 mm × 25 mm) at the C1–C2 level. The lesion was hypointense on T1- and hyperintense on T2-weighted sequences, and intensely enhanced with contrast, but showed no “dural tail” (i.e., as typically seen with meningiomas) [Figures 1 and 2]. Through a C1 laminectomy, and restricted posterior foramen magnum osteotomy, she underwent gross total microsurgical resection of a well-demarcated lesion. At surgery, the tumor was subdural, but showed no dural attachment. Rather, it was adherent both to the spinal accessory nerve and vertebral artery (i.e., laterally and rostrally).

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.

©2022 Published by Scientific Scholar on behalf of Surgical Neurology International

Table 1: Summary of previously reported spinal meningiomas without dural attachment.

Authors	Ko <i>et al.</i> ^[3] 2011	Maamri <i>et al.</i> ^[4] 2022	Mohri <i>et al.</i> ^[5] 2019	Zhang <i>et al.</i> ^[7] 2021
Patient's age (y.o.), sex	34, woman	58, woman	69, woman	45, woman
Level	L2-3	L3	Cranio cervical junction	L3
Origin	Nerve root	Nerve root	Spinal accessory nerve	Nerve root
Degree of resection	Total	Total	Total	Total
Histological subtype	Clear cell	Clear cell	Meningothelial	Clear cell
Outcome	Good	Good	Good	Good

Number in the parentheses indicates reference numbers in the manuscript

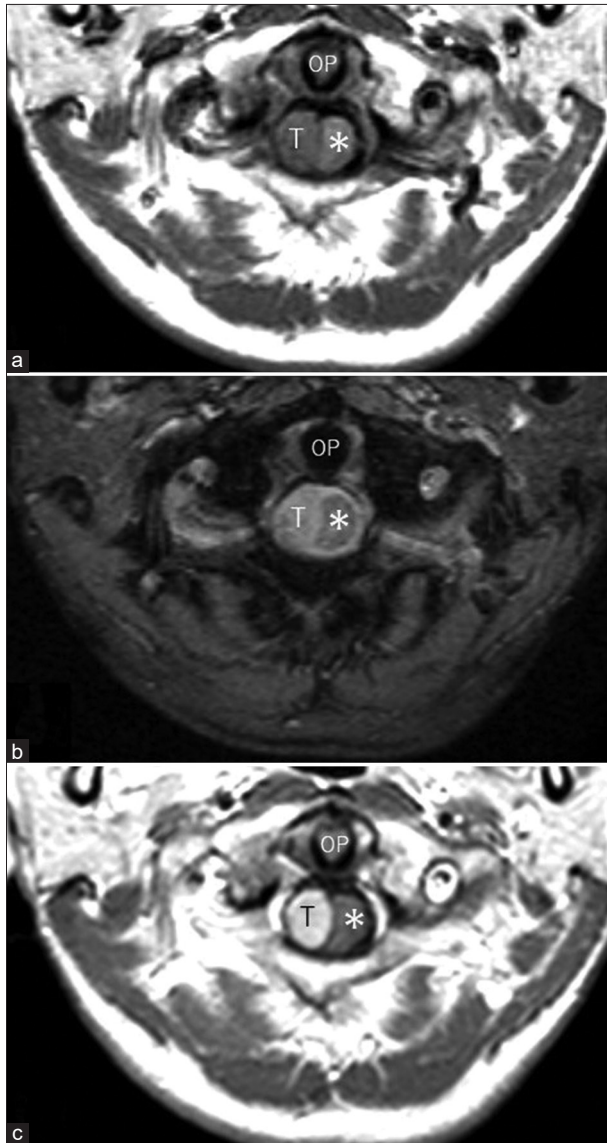


Figure 1: Axial T1- (a), T2- (b), and post-contrast axial (c) T1-weighted magnetic resonance images at the same level showing a well-demarcated extramedullary tumor (T) compressing the spinal cord from the right lateral side (asterisk). The tumor appears hypointense on T1- and hyperintense on T2-weighted sequences relative to the spinal cord, and intensely enhanced. OP: Odontoid process.

Pathology

The following pathological features were consistent with an atypical meningioma; anaplastic cells with short spindle-shaped nuclei and an intervening rich vasculature, four mitotic figures per 10 high-power fields; and scattered necrotic foci [Figure 3].

Immunohistochemistry

The immunohistochemical examination was consistent with an atypical meningioma showing; positive staining for epithelial membrane antigen and progesterone receptors; and negative staining for glial fibrillary acidic protein. The Mindbomb Homolog-1 index was 10% [Figure 4].

Postoperative course

The postoperative course was uneventful as the patient remained neurologically intact. Although she received no prophylactic postoperative radiotherapy, she remains under close surveillance with MRI scans repeated every 6 months looking for tumor recurrence.

DISCUSSION

Original of spinal meningiomas

Most spinal meningiomas develop as dural-based and intradural extramedullary tumors.^[1,6] In the present case, as we observed no dural attachment, we assumed that the tumor originated from the spinal accessory nerve. We identified four similar cases of spinal meningiomas originating from the spinal accessory nerve; histologically, three were meningothelial, and one was psammomatous.^[3-5,7]

Recommendation for gross total excision for atypical spinal meningiomas to achieve the best long-term prognosis

Spinal atypical meningiomas are typically aggressive lesions with a poor prognosis, and, therefore, warrant aggressive treatment.^[2] For these lesions, it is critical to plan timely surgery for gross total resection that is possible without major neurological sequelae.^[1,6] Certainly, the early surgery before



Figure 2: Post-contrast coronal (a) and midsagittal (b) T1-weighted magnetic resonance images showing an oval tumor at the C1–C2 levels. It measures 12 × 10 × 25 mm in diameter and had no dural tail. C1: Atlas; C2: Axis; T: Tumor.

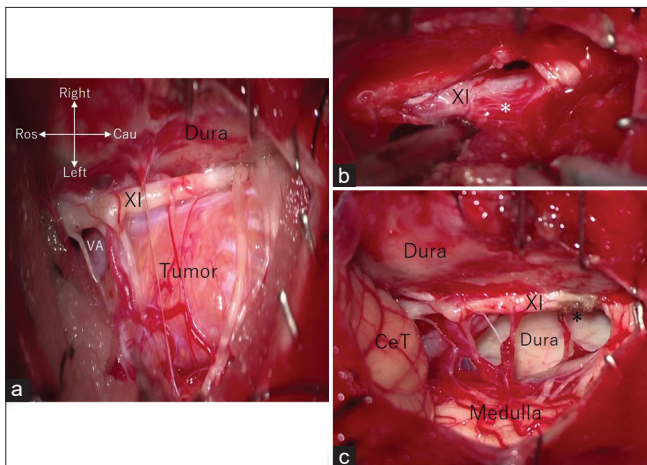


Figure 3: Intraoperative photos showing (a) reflection of the dura mater exposing the tumor adjacent to the right spinal accessory nerve (XI) and vertebral artery (VA), (b) tumor adhesion (asterisk) to the right spinal accessory nerve (XI) exposed following partial tumor resection, and (c) post-total resection with the intact medulla, spinal accessory nerve (XI), tumor attachment site (asterisk), and the surrounding dura mater. Cau: Caudal; CeT: Cerebellar tonsil; Ros: Rostral.

the onset of significant neurological deficits is associated with the best long-term prognoses for these lesions.^[1] In our case,

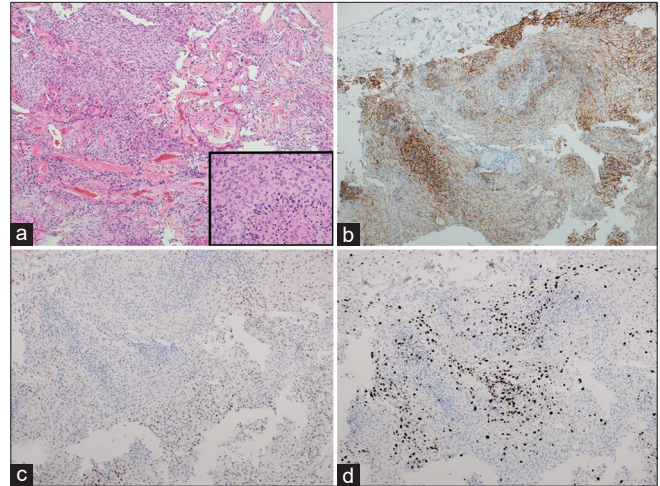


Figure 4: Photomicrographs of the resected specimens showing tumor tissue comprised short spindle-shaped nuclei and intervening vasculature. The magnified view of the tumor cells (a), immunohistochemical analysis showed positive staining for epithelial membrane antigen (b), and progesterone receptor (c). The Mindbomb Homolog-1 index accounted for 10% (d). (a) Hematoxylin and eosin stain, ×100; inset in (a), ×400; (b-d) ×100.

immediate radiotherapy was not administered because the tumor was totally resected and was only focally adherent to the spinal accessory nerve, not the dura.

CONCLUSION

Atypical meningiomas can originate from the spinal accessory nerve. Timely, surgery with gross total removal results in the best long-term outcomes.

Declaration of patient consent

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

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Bayoumi AB, Laviv Y, Karaali CN, Ertlav K, Kepoglu U, Toktas ZO, et al. Spinal meningiomas: 61 cases with predictors of early postoperative surgical outcomes. *J Neurosurg Sci* 2020;64:446-51.
2. Kawaguchi Y, Ishihara H, Abe Y, Seki S, Tokunaga A, Urushizaki A, et al. Fatal prognosis of an atypical meningioma in the cervical spine. *J Orthop Sci* 2008;13:155-9.

3. Ko JK, Choi BK, Cho WH, Choi CH. Non-dura based intraspinal clear cell meningioma. *J Korean Neurosurg Soc* 2011;49:71-4.
4. Maamri K, Taieb MA, Trifa A, Elkahla G, Njima M, Darmoul M. Spinal clear cell meningioma without dural attachment: A case report and literature review. *Radiol Case Rep* 2022;17:1760-4.
5. Mohri M, Yamano J, Saito K, Nakada M. Spinal accessory nerve meningioma at the foramen magnum with medullary compression: A case report and literature review. *World Neurosurg* 2019;128:158-61.
6. Roux FX, Nataf F, Pinaudeau M, Borne G, Devaux B, Meder JF. Intraspinal meningiomas: Review of 54 cases with discussion of poor prognosis factors and modern therapeutic management. *Surg Neurol* 1996;46:458-64.
7. Zhang X, Zhang P, Wang JJ, Dong S, Wu Y, Zhang H, *et al.* Intraspinal clear cell meningioma without dural attachment: A case report and literature review. *Medicine (Baltimore)* 2021;100:e25167.

How to cite this article: Ueno H, Tsutsumi S, Hashizume A, Sugiyama N, Ishii H. Atypical meningioma originating from the spinal accessory nerve. *Surg Neurol Int* 2022;13:598.

Disclaimer

The views and opinions expressed in this article are those of the authors and do not necessarily reflect the official policy or position of the Journal or its management. The information contained in this article should not be considered to be medical advice; patients should consult their own physicians for advice as to their specific medical needs.