

CASE REPORT

An unexpected complication of hypoglossal schwannoma surgery: Cerebrospinal fluid leakage—A case report and literature review

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Abstract

Background: Extracranial hypoglossal schwannoma is a rare tumor primarily treated with surgical excision. This article aims to highlight the potential for unexpected complications intraoperatively, such as cerebrospinal fluid leakage from skullbase to neck.

Methods: A previously healthy 23-year-old male presented with tongue numbness. Magnetic resonance imaging revealed a 17 × 20 mm nodular lesion adjacent to the cervical segment of the internal carotid artery. Surgical excision was scheduled due to suspicion of a neurogenic tumor.

Results: Intraoperatively, despite careful handling, cerebrospinal fluid leakage was observed. Manipulation of the mass caused detachment of proximal nerve fibers, potentially indicating avulsion of the hypoglossal nerve from the brainstem or nearby. Clear fluid leakage from the skull base was also noted.

Conclusion: Thorough preoperative evaluation and patient education regarding potential complications are crucial. This article presents an unexpected complication encountered during surgical excision of extracranial hypoglossal schwannoma, emphasizing the need for awareness and preparedness in such cases.

KEYWORDS

avulsion, cerebrospinal fluid leakage, hypoglossal schwannoma, neck tumor, surgery

1 | INTRODUCTION

The hypoglossal nerve is the twelfth cranial nerve that innervates the muscles of the tongue and provides branches to the cervical plexus to assist in innervating the muscles of the cervical region.¹ Hypoglossal schwannoma is a tumor that accounts for approximately 5% of head and neck schwannomas and is composed entirely of motor fibers.² This tumor can be entirely intracranial, intracranial/extracranial, or solely extracranial. Although the extracranial origin is the rarest type, tumors in this area are more commonly observed in the parapharyngeal

and sublingual regions and can mimic paragangliomas.^{2,3} While tongue wasting is the most common finding in hypoglossal schwannomas, extracranial tumors can present with a neck mass.⁴ Magnetic resonance imaging (MRI) holds a significant place among radiological imaging modalities in the evaluation of hypoglossal schwannomas with low signal intensity on T1-weighted imaging and high intensity imaging on T2-weighted imaging.⁵ The recommended treatment for extracranial schwannomas is generally surgical excision; however, preserving nerve function can be quite challenging.⁶ In this article, we aim to present an unexpected complication that we

encountered during surgical excision in a patient with hypoglossal schwannoma.

2 | CASE REPORT

A 23-year-old male patient with no previously known illness presented to our clinic with complaints of numbness in the tongue, headache, and occasional dizziness for 3 years. Physical examination revealed normal cranial nerves and no palpable neck masses. Despite inconclusive findings from patient's medical history-based tests, ultrasonography showed a well-defined, hypoechoic solid lesion located deeply adjacent to the left parotid gland. Contrast enhanced CT of neck revealed a mass lesion adjacent to the cervical segment of the left internal carotid artery, measuring 20×15 mm, with contrast enhancement observed within vascular structures. Subsequent magnetic resonance imaging (MRI) revealed a nodular lesion measuring 17×20 mm in the left parapharyngeal area adjacent to the cervical segment of the internal carotid artery, showing a heterogeneous-hyperintense signal on T2-weighted images and marked heterogeneous contrast enhancement after intravenous administration of contrast agent (Figure 1). Cervical MR angiography showed normal contour and patency of both common carotid arteries and internal carotid arteries in the imaged sections. Fine needle aspiration biopsy was not performed due to the proximity of the mass to vital structures. Neurogenic tumor was the primary suspected diagnosis and the patient was scheduled for surgical excision. Informed consent was obtained and total

tumor excision was planned. The skin, subcutaneous tissue, and platysma were passed through by making a transverse cervical incision. Dissection was performed anteriorly to the sternocleidomastoid muscle. The internal jugular vein and carotid artery were visualized. Several lymph nodes of undetermined significance were excised at Level 2. Then, the fascia overlying the submandibular gland was incised and the digastric muscle was followed up to the mastoid tip. A mass lesion suspected to be a schwannoma originating from the hypoglossal nerve was observed in the deep portion of the digastric muscle. The mass had well-defined borders and continuous with the hypoglossal nerve. The distal portion of the hypoglossal nerve was transected close to the mass with enough surgical margin, after it was confirmed that the mass was originating from the nerve. The mass was dissected and separated from the surrounding tissues. To visualize the proximal portion of the mass, the digastric muscle was incised. The proximal portion of the nerve was followed up to the skull base. It was observed that the mass was located near the hypoglossal canal in a manner that would not allow for meticulous dissection. During digital manipulation of the proximal segment of the mass, detachment in the proximal portion of the nerve fibers was observed (Figure 2), which is thought to be an avulsion of the hypoglossal nerve from the brainstem or somewhere between the brainstem and hypoglossal canal. Additionally, clear fluid leakage from the skull base was observed. Initially, the precise nature of the fluid could not be determined. Subsequently, it was observed that the fluid continued to leak in a manner of a leakage

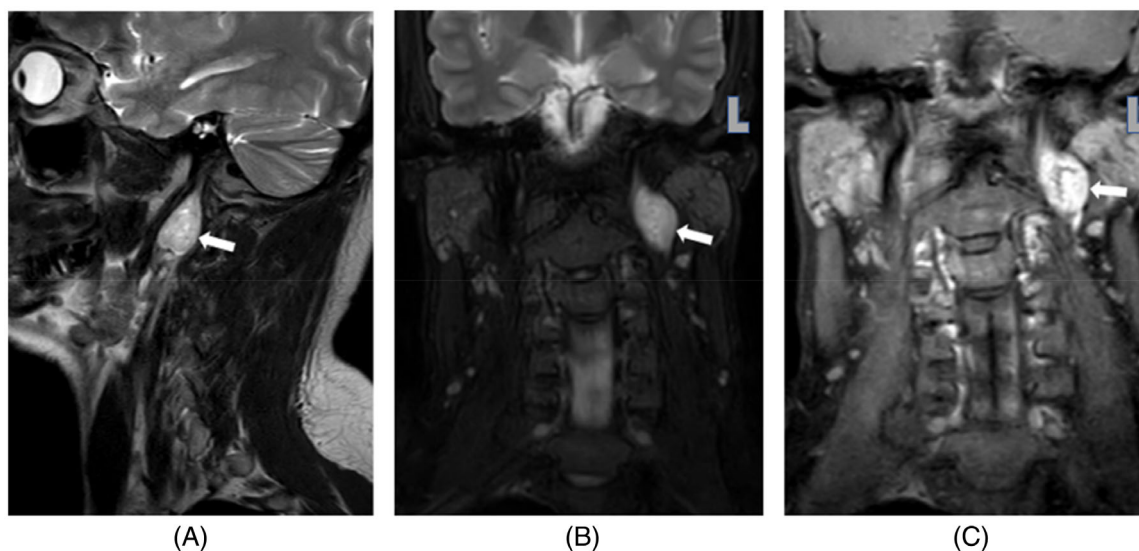


FIGURE 1 MRI findings. (A) Sagittal T2-WI showed a mass with heterogeneous signal hyperintensity (arrow). (B) Coronal T2-WI (arrow). (C) Contrast-enhanced T1-WI showed a mass with heterogeneous enhancement on coronal section (arrow) [Color figure can be viewed at wileyonlinelibrary.com]



FIGURE 2 Surgical specimen. Proximal end of hypoglossal nerve (star). Distal end of hypoglossal nerve (arrow). The location where the mass abuts the hypoglossal canal (arrowhead). The intracranial segment of hypoglossal nerve is approximately 3 cm in length (bidirectional arrow) [Color figure can be viewed at wileyonlinelibrary.com]

after aspiration. When the anesthesiologist performed maneuvers to increase intracranial pressure, an increased amount of fluid was seen. It was determined that the fluid was cerebrospinal fluid. The defect site of cerebrospinal fluid (CSF) leakage was repaired using a muscle graft harvested from the sternocleidomastoid muscle and sealed with Surgicel® and Floseal®. The patient was monitored until CSF leakage completely ceased. After careful evaluation, a mini-vac drain was placed in the surgical field following hemostasis. The surgical field was closed with appropriate suturing and mini-vac drain was placed. The postoperative course was uneventful and no neural deficit except for unilateral hypoglossal palsy was observed. The fluid coming from the drain appeared serosanguineous on the first postoperative day, and then decreased in volume and appeared clear in color in the following days. On the third postoperative day, the drain was removed. To reduce cerebrospinal fluid leakage, pressure dressing was applied on the patient's neck. Complications such as headaches, neck pain, seizure, and meningitis that may be seen due to cerebrospinal fluid leakage were not observed. Histopathologic evaluation of surgical specimen

confirmed hypoglossal schwannoma. The patient was discharged home on postoperative day 5. Regular outpatient follow-up visits, including clinical examinations and cervical ultrasound every 6 months, were scheduled.

3 | DISCUSSION

Hypoglossal schwannoma is a rare tumor among extracranial head and neck schwannomas, particularly when it originates from the descending loop of the hypoglossal nerve.^{5,7,8} The current literature recommends surgical excision as the first-line treatment for schwannoma. For Schwannomas arising from other cranial nerves, excision of the mass en bloc with the entire nerve is the best option in order to achieve a curative treatment.⁹ It has been suggested that en bloc resection of the nerve reduces the risk of recurrence, the need for repeat surgery, and the potential for sarcomatous transformation.^{7,10} However, in another study, intracapsular enucleation was shown to preserve nerve function and no recurrence was seen in patients.¹¹ Our approach was to perform en bloc resection of the tumor and its origin tissue. No postoperative complications were observed except for hypoglossal nerve paralysis on the surgical side. However, despite our meticulous and careful approach, CSF leakage was observed intraoperatively. The CSF leakage was primarily caused by difficulty in dissecting the mass at the hypoglossal canal level and by avulsion of the proximal nerve fibers during digital manipulation. There are studies reporting intraoperative CSF leakage, particularly in Jugular Foramen Schwannomas.¹² Similarly, cases of CSF leakage after hypoglossal schwannoma surgery have been reported. Suri et al. reported a CSF leak in one of 12 patients with intradural located hypoglossal schwannoma after surgery.⁴ In the study conducted by Sarma et al., it was demonstrated that one out of three patients with intracranial hypoglossal schwannoma experienced CSF leakage following surgery.¹³ To our knowledge, this is the first reported case demonstrating the possibility of intraoperative CSF leakage from the skull base to the neck in a patient with extracranial hypoglossal schwannoma. Surgical teams should be aware that, despite meticulous and careful surgical approaches, rare complications such as CSF leakage may occur in patients with masses extending into hypoglossal canal, and should be prepared to manage such complications.

4 | CONCLUSION

Extracranial hypoglossal schwannoma is a rare tumor and its treatment is generally surgical excision. Prior to

surgery, a detailed workup of the patient should be conducted and the patient should be informed about possible situations that may arise. In this article, we aim to emphasize the possibility of unexpected complications such as CSF leakage during surgery.

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CONFLICT OF INTEREST STATEMENT

The authors declare no conflicts of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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