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Case Report

Successful treatment of recurrent extracranial hypoglossal schwannoma using the neuroendoscopic transnasal far-medial approach

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ABSTRACT

Background: Extracranial hypoglossal schwannomas are rare, and transcranial skull base surgery can be challenging due to their proximity to the lower cranial nerves, jugular vein, vertebral artery, and carotid artery. The application of neuroendoscopic surgery for extracranial hypoglossal schwannomas has rarely been reported.

Case Description: A 53-year-old woman previously underwent lateral suboccipital surgery for a hypoglossal schwannoma when she was 25 years old. The patient had experienced aggravated dysphagia over the past month. Radiological examination revealed a recurrent extracranial hypoglossal schwannoma invading the left side of the clivus. The neuroendoscopic transnasal far-medial approach was performed, and the recurrent schwannoma was completely removed without any significant perioperative complications or recurrence for 3 years.

Conclusion: Our report highlights the usefulness of the neuroendoscopic transnasal far-medial approach for the removal of recurrent extracranial hypoglossal schwannomas. The neuroendoscopic approach offers a viable and less invasive alternative to traditional transcranial skull-base surgery, especially in complex cases involving critical anatomical structures. The reported case study underscores the potential of neuroendoscopic surgery as a valuable tool in managing challenging skull-base tumors.

Keywords: Extracranial hypoglossal schwannoma, Neuroendoscopic transnasal far-medial approach, Skull-base surgery

INTRODUCTION

Hypoglossal schwannomas are rare, benign tumors that can grow along the tract of the hypoglossal nerve; however, it is quite rare to see their extracranial extension.[11] The clinical manifestations of hypoglossal schwannomas vary according to the extent of the tumor and the adjacent nerves. The representative symptoms include dysphagia, dysarthria, headache, and tongue atrophy. [2,4,5,11] Surgical resection is a well-established management strategy for symptomatic hypoglossal schwannomas. A transcranial skull base approach is often performed to remove hypoglossal schwannomas. [11] The use of neuroendoscopic surgery to remove hypoglossal schwannomas has rarely been reported.

However, transnasal or transoral neuroendoscopic surgery is currently utilized to approach lower clival and petroclival regions safely. [12] Furthermore, the neuroendoscopic transnasal far-medial

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approach can help surgeons confirm lateral anatomical structures to the midline. [9] Using this approach, the condylar, jugular tubercle and hypoglossal canal can be visualized, allowing for the resection of tumors such as chordomas, chondrosarcomas, osteochondromas, and meningiomas. [1,3,8,10] However, to the best of our knowledge, the neuroendoscopic transnasal far-medial approach for removing hypoglossal schwannomas has not yet been described.

In this study, we present a case of a recurrent extracranial hypoglossal schwannoma that was successfully treated with neuroendoscopic surgery, highlighting the effectivity of the transnasal far-medial approach.

CASE PRESENTATION

A 53-year-old woman presented with complaints of hoarseness and dysphagia that had been gradually aggravating over the past 3 months. At the age of 25 years, she underwent surgery to remove a left dumbbell-type

hypoglossal schwannoma, which resulted in atrophy of the left tongue. Following the initial surgery, she experienced residual tongue atrophy, temporary facial nerve palsy, slight left vocal cord palsy, and dysphagia. Despite these challenges, she remained independent in her daily activities. She continued to attend follow-up appointments at our outpatient clinic until she ceased visiting at the age of 46 years. The patient had experienced aggravated dysphagia in the past month. No significant changes were observed in residual left tongue atrophy, left vocal cord palsy, and soft palate asymmetry. However, a swallowing endoscopy revealed residual saliva in the radix linguae and piriform sinuses. Due to dysphagia, the patient was unable to swallow the general diet. Magnetic resonance imaging revealed a cystic tumor lesion on the left side of the clivus, indicating a residual extracranial hypoglossal schwannoma invading the clivus and compressing the brainstem [Figure 1]. Surgery for tumor removal using a neuroendoscope was planned to alleviate these symptoms.

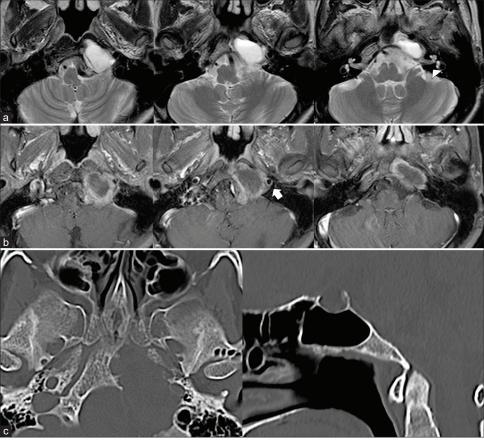


Figure 1: Preoperative head magnetic resonance (a: T2-weighted images and b: gadolinium-enhanced T1-weighted images) and computed tomography images (c: left, axial images; right, sagittal images). (a and b) The cystic tumor tumoral lesion is located on the left side of the clivus, showing ring enhancement with gadolinium contrast. The lesion extends into the intracranial space, compressing the brain stem and left internal carotid artery (white arrowhead: internal auditory canal, white arrow: internal carotid artery). (c) The tumor has invaded the left side of the clivus.

The patient was sedated under general anesthesia and subjected to the neuroendoscopic endonasal far-medial approach to access the tumor lesion. The anterior wall of the sphenoid sinus was approached through the left nasal cavity. The mucosa on the anterior wall of the sphenoid sinus and epipharynx was peeled to expose the sphenoid bone and clivus, avoiding arterial injury guided by intraoperative navigation and Doppler ultrasonography [Figure 2a]. Subsequently, a tumor lesion posteriorly invading the left eustachian tube was identified. The anterior walls of the sphenoid bone and clivus were drilled, and the superior and medial sides of the tumor lesion were confirmed [Figure 2b]. Under a neuroendoscope angled at 30°, the tumor was detached along the margin between the capsule and the clivus. Subsequently, the dura of the petrous bone was identified. The tumor capsule was gradually detached from the posterior, lateral, and anterior surfaces of the petrous bone, and the soft tumor lesion was removed using suction. The dura was intact, suggesting that the tumor had grown from the extracranial space [Figure 2c]. A small amount of cerebrospinal fluid (CSF) leaked after removing the tumor from the hypoglossal canal [Figure 2d]. We hypothesized that the residual extracranial hypoglossal schwannoma

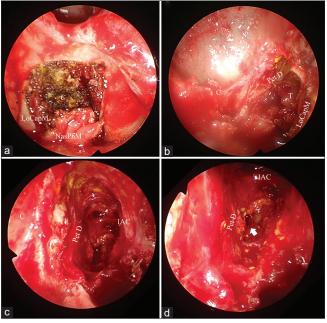


Figure 2: Intraoperative neuroendoscopic findings. (a) The clivus becomes visible after the nasal pharyngeal membrane is detached caudally. (b) The yellowish tumor lesion is identified lateral to the clivus and dorsal to the longus capitis muscle. (c and d) Views using a neuroendoscope angled at 30°. After subtotal resection of the tumor, the dura of the petrous bone and the inner wall of the internal auditory canal are exposed. The tumor invading the hypoglossal canal (white arrow) is also removed. (C: Clivus; IAC: Internal auditory canal; LoCapM: Longus capitis muscle; NasPhM: Nasal pharyngeal membrane; PetD: Dura of the petrous bone; T: Tumor).

extended intracranially through the hypoglossal canal and compressed the brainstem. The tumor capsule was visually identified and resected, and the removal procedure was completed. The dural surface was then covered with Duragen (Integra Life Sciences, NJ, USA), and the removal cavity was filled with abdominal fat tissue, Surgicel (Ethicon, Johnson and Johnson Medical Ltd., Tokyo, Japan), and fibrin glue. The mucosal flap was then sutured and covered with fibrin glue. In the absence of any evident persistent bleeding or CSF leakage, the operation was terminated. Intraoperative and postoperative pathological diagnoses were consistent with a schwannoma. The postoperative course was uneventful, and no complications, such as CSF leakage, occurred. The preoperative dysphagia and hoarseness disappeared after surgery, and the patient was able to consume a general diet. The patient was discharged from the hospital 12 days after the surgery. Follow-up radiological examination revealed gross total removal of the recurrent schwannoma [Figure 3]. The patient is currently being followed up in an outpatient clinic, and the tumor has not recurred 3 years after the surgery.

DISCUSSION

Herein, we describe a case of a recurrent hypoglossal nerve schwannoma that was successfully treated using the neuroendoscopic endonasal far-medial approach. The schwannoma was completely resected, and no postoperative complications occurred.

Hypoglossal schwannomas are generally treated with craniotomy; however, this surgical approach has the disadvantages of high mortality and morbidity. With the development of surgical techniques and microanatomical knowledge, safe and effective craniotomies have been performed.[11] Traditional craniotomy surgery involves a lateral or midline suboccipital approach, which is only suitable for intracranial hypoglossal schwannomas without extension into the hypoglossal canal. Far-lateral, supracondylar, transcondylar, or transcondylar fossa approaches can be applied to extra- and intracranial hypoglossal schwannomas. Among these approaches, the extreme lateral infrajugular transcondylar-transtubercular exposure (so-called "ELITE") approach has been advocated.[11] This technique includes an extended lateral suboccipital craniotomy, mastoidectomy, partial occipital condyle and jugular tubercle removal, sigmoid sinus, and jugular bulb skeletonization, and vertebral artery decompression with or without mobilization. This method enables access to extra- and intracranial hypoglossal schwannomas. However, craniotomy has limitations in terms of its invasiveness.

Neuroendoscopic transoral or transnasal surgery is less invasive for skull-base tumors.[6,7,13,14] In the present case study, the neuroendoscopic endonasal far-medial approach was used to resect a recurrent extracranial hypoglossal

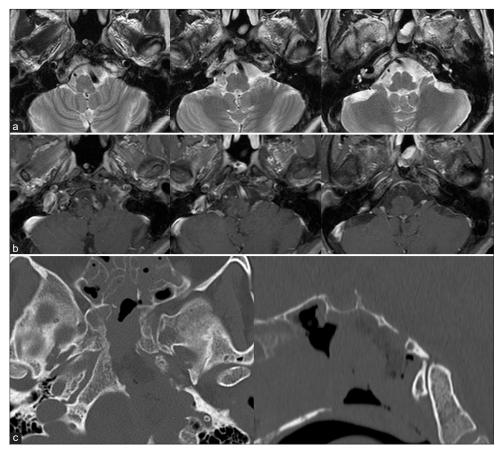


Figure 3: Postoperative head magnetic resonance (a: T2-weighted images and b: gadoliniumenhanced T1-weighted images) and computed tomography images (c: left, axial images; right, sagittal images). (a and b) Cystic and ring-enhanced tumors are completely removed. Brainstem compression by the tumor is also resolved. (c) The access route from the sphenoid sinus to the clivus is observed.

schwannoma. Ali et al. described the essential steps of this approach: (a) opening the sphenoid sinus and identifying the paraclival internal carotid artery; (b) making an inverted U-shaped flap of the nasal pharyngeal membrane, exposing the supracondylar groove and lower clivus; (c) identifying the pathway of the internal carotid artery using Doppler ultrasonography and an intraoperative navigation system; and (d) drilling the lower clivus from the midline to the lateral side and confirming the hypoglossal canal, jugular tubercle, and occipital condyle.[1] However, in our case, the hypoglossal schwannoma invaded the clivus and petrous bone and was observed lateral to the clivus. The important anatomical structures, including the jugular tubercle and occipital condyle, were not intraoperatively identified. Therefore, using an intraoperative navigation system, by following the lateral margin between the tumor capsule and clivus, we removed the tumor from the hypoglossal canal. While identifying anatomical landmarks intraoperatively is important, intraoperative navigation systems can also be useful in instances where tumoral bony invasion results in the loss of usual anatomical structures.

Neuroendoscopic surgery is less invasive than craniotomies as the internal jugular vein and lower cranial nerves do not have to be exposed; this is one advantage of neuroendoscopic surgery. Unlike previous tumors growing from the midline anatomical structures, such as the clivus or its dura, the neuroendoscopic endonasal far-medial approach can be helpful for treating hypoglossal schwannomas growing from the hypoglossal canal. [1,3,8,10] Regarding the pitfalls related to the neuroendoscopic endonasal far-medial approach, the following adverse events have been noted: injury of the clival internal carotid artery, basilar artery, or vertebral artery; abducent nerve palsy; CSF leakage; and craniocervical instability due to excessive occipital condyle drilling.^[1] CSF leakage was also observed in our case. However, appropriate management resolves these complications.

CONCLUSION

We report a case of recurrent hypoglossal schwannoma that was successfully treated using the neuroendoscopic transnasal far-medial approach. As neuroendoscopic surgery is less invasive than craniotomy, recurrent extracranial hypoglossal schwannoma can be approached with this surgical method. However, the usefulness of this method should be evaluated in further similar cases.

Ethical approval

The research/study approved by the Institutional Review Board at Medical Research Institute KITANO HOSPITAL, PIIF Tazuke-kofukai, number P221000400, dated October 17, 2022.

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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