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Meningiomas Involving the Hypoglossal Canal: A Case Report and Literature Review

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Abstract

We report a rare case of hypoglossal canal meningioma in a 65-year-old woman who presented with dysphagia. Neurological examination revealed left hypoglossal nerve palsy. Head computed tomography and magnetic resonance imaging revealed a lesion around the left hypoglossal canal. She underwent a total resection with a midline suboccipital transcondylar approach. There were no postoperative complications, and the hypoglossal nerve palsy improved. There was no recurrence nine months after the surgery. Choosing a surgical approach that considers the site of origin and extent of tumor extension is important.

Keywords: hypoglossal canal, hypoglossal nerve, meningioma, midline suboccipital transcondylar approach, transcondylar approach

Introduction

Meningiomas are common intracranial neoplasms and represent 19%-35% of all primary intracranial tumors.[1,2] Hypoglossal canal meningiomas are rare, with five cases reported to date.[2-6] We report a case of hypoglossal canal meningioma with small intracranial extension and review the literature.

Case Report

For two months, a 65-year-old woman reported difficulty swallowing and speaking (left hypoglossal nerve paralysis). Neurological examination revealed tongue deviation to the left due to left hypoglossal nerve impairment.

Magnetic resonance imaging (MRI) showed an enhanced mass extending intracranially from the left hypoglossal canal (10 mm × 7.5 mm) (Fig. 1A). Computed tomography (CT) showed calcification of the tumor (Fig. 1B). The lesion did not dilate the hypoglossal canal (Fig. 1C).

Tumor removal was performed via the transcondylar approach using midline suboccipital craniotomy (Fig. 2). The navigation system and hypoglossal nerve monitoring by direct stimulation were performed with the patient in a prone position. A midline linear vertical skin incision was made from the inion to the C4 spinous process. The muscles were divided bilaterally, and the suboccipital space was exposed. A right-side-dominant midline suboccipital craniotomy was performed, and the foramen magnum was opened. The condyle was drilled, and the hypoglossal canal was opened (Fig. 2A). The dural incision was made in a Y-shaped fashion. The tumor was yellowish and soft (Fig. 2B and C). The tumor was completely resected microscopically with dural attachment coagulation (Simpson grade II) (Fig. 2D). Based on the dural attachment localization, the tumor origin seemed to be in the hypoglossal canal, from which it extended intracranially. The hypoglossal nerve was morphologically preserved, and monopolar nerve stimulation could not detect hypoglossal nerve electromyography waves on the left side. Pathological examination showed that the histological subtype was meningothelial meningioma with a MIB-1 index of 3% (Supplementary Fig. 1).

There were no postoperative complications. The postoperative head MRI showed total tumor removal (Fig. 3A), and the postoperative CT bone scan confirmed that the

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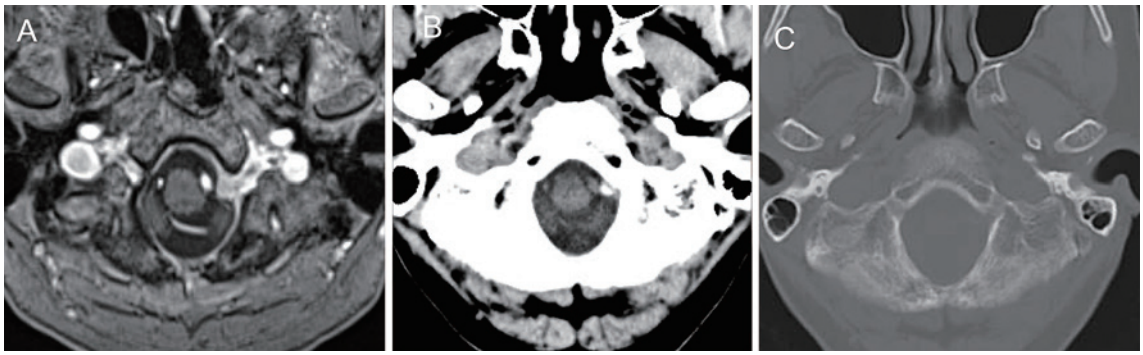

Fig. 1
(A) Preoperative head MRI shows a mass extending intracranially from the left hypoglossal canal.
(B) Preoperative head CT shows calcification of the tumor.
(C) Bone scan shows no destruction or enlargement of the hypoglossal canal.

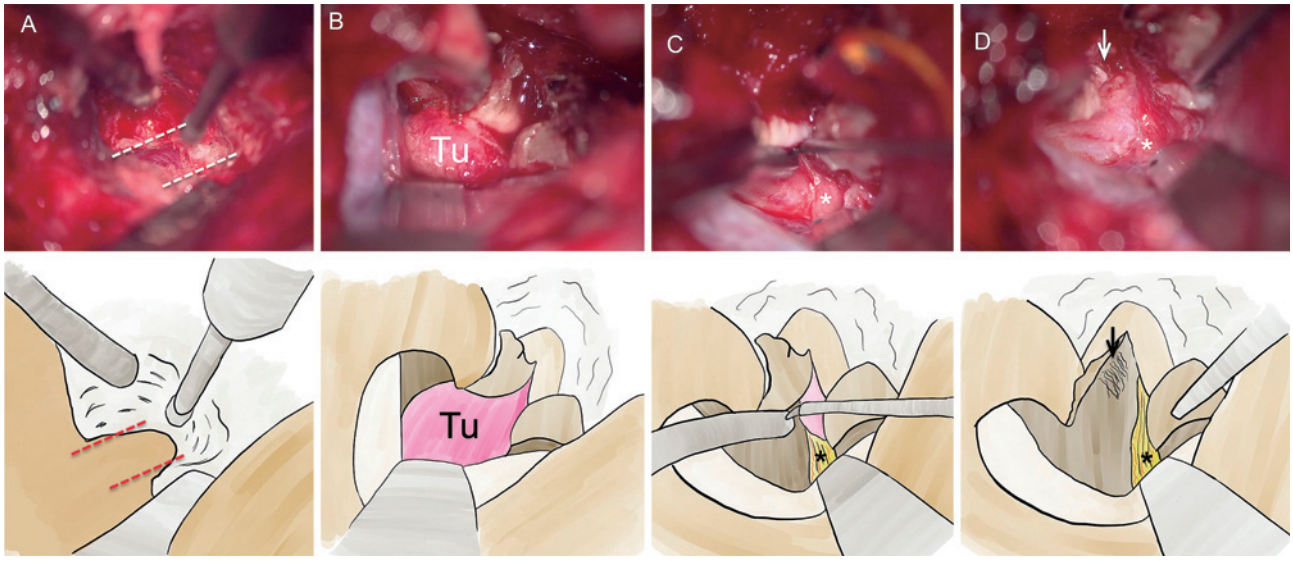

Fig. 2
Intraoperative findings. The upper row shows the intraoperative photographs, and the lower row shows the corresponding illustrations. (A) The hypoglossal canal was opened. (B) After a dural incision, a yellowish and soft tumor was identified. (C) The tumor was detached from the hypoglossal nerve. (D) The tumor was completely removed.
Dotted line: hypoglossal canal, asterisk: hypoglossal nerve, arrow: tumor attachment
Tu: tumor

condylar resection had been performed as planned (Fig. 3B and C). Preoperative hypoglossal nerve impairment improved on the first postoperative day after surgery. There was no tumor recurrence nine months after the operation.

Discussion

Review of hypoglossal canal meningioma

Meningiomas are relatively common intracranial tumors. However, meningiomas arising from the hypoglossal canal are very rare. Foramen magnum meningiomas are tumors that arise anteriorly from the lower one third of the clivus to the superior border of the C2 body, laterally from the jugular tubercle to the superior border of the C2 lamina, and posteriorly from the anterior border of the occipital scales to the C2 spinous process. Although hypoglossal canal meningiomas are also foramen magnum meningiomas, they involve the hypoglossal canal. Calzada et al. published the first report of a meningioma in the hypoglossal canal in 2007, and five cases have been reported since then.[2-6] Table 1 shows the reported cases of hypoglossal canal meningioma to date, including our case.

The frequency of laterality was the same for both the left and right sides. There were four females and two males. The mean age of the patients was 52.6 years. Initial symptoms include dysarthria, dysphagia, hoarseness, snor-

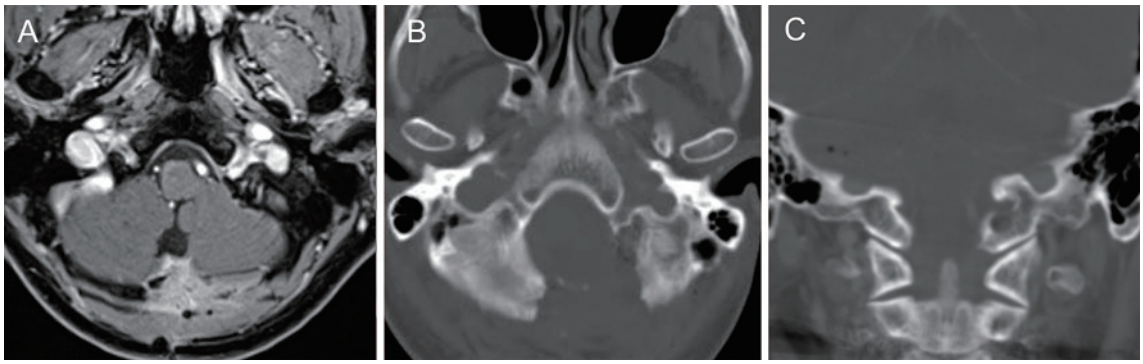

Fig. 3
(A) Postoperative head MRI shows total removal of the tumor.
(B) Postoperative head CT bone scan (axial view) shows resection of the condyle.
(C) Postoperative head CT bone scan (coronal view) shows preservation of the occipitoatlantal joint.

ing, headache, lightheadedness, and neck mass in the order of frequency. Most neurological findings included hypoglossal nerve palsy, tongue atrophy, and vocal cord palsy.

Depending on the localization of the tumor, the tumor can be classified into four patterns (Supplementary Fig. 2). Localization includes the hypoglossal canal with intracranial or extracranial localization. The combination of these determines the following classification. The hypoglossal canal only (canal type) (Supplementary Fig. 2A), hypoglossal canal-intracranial (cisternal type) (Supplementary Fig. 2B), hypoglossal canal-extracranial (extracranial type) (Supplementary Fig. 2C), and intracranial-hypoglossal canal-extracranial (dumbbell type) (Supplementary Fig. 2D). In the case of the neck, the differential includes carotid body tumors and glomus tumors.[2)]

The surgical approach was selected according to the lesion removed in each localization. The transcondylar approach for canal type, midline suboccipital, and far lateral approach for cisternal type and transcervical approach for extracisternal and dumbbell types have been reported. In terms of surgical results, the degree of removal was 66.7% (4/6) for total gross removal and 33.3% (2/6) for subtotal removal (STR). STR has been reported in dumbbell-type[4)] and in a case with large intracranial lesions.[3)] The hypoglossal nerve was preserved in three cases, with a preservation rate of 60% (3/5, hypoglossal nerve preservation was not reported in one case).

Postoperative complications were described in five cases, with a complication rate of 20% (1/5); pseudomeningocele was observed. Furthermore, the outcome of hypoglossal nerve palsy was described in three cases, with 66.7% (2/3) showing an improvement and 33.3% (1/3) worsening. The mean follow-up period was 32.3 months.

Surgical consideration

The operative indication is the prevention of destruction of the craniovertebral junction and relieving compression of surrounding tissues such as the brain stem and lower cranial nerves.[6)] Improving hypoglossal nerve impairment is also an important treatment objective.

The surgical approach depends on the localization of the tumor (Supplementary Fig. 2) and the treatment objective. The transcervical approach is selected if the tumor extends extracranially into the neck (extracranial type) (Supplementary Fig. 2C), a transcondylar approach if the tumor is localized to the hypoglossal canal (canal type) (Supplementary Fig. 2A), and a midline suboccipital, and far lateral approach if the tumor extends intracranially from the hypoglossal canal (cisternal type) (Supplementary Fig. 2B). For dumbbell type, different approaches should be used depending on the purpose (Supplementary Fig. 2D). If the extracranial lesion is to be removed, the transcervical approach should be selected. If the intracranial lesion is to be removed, a midline suboccipital and far lateral approach should be selected; and if both are to be removed, a combination of these approaches should be selected. If the tumor extends from the hypoglossal canal to the intracranial space and decompression of the hypoglossal nerve is intended, as in our case, midline suboccipital craniotomy with condyle resection (midline suboccipital transcondylar approach) should be performed to bring the hypoglossal canal to the intracranial space within the same field of view. Although approaches without condyle resection can preserve condyle stability, it is difficult to remove tumors in the hypoglossal canal. On the other hand, although approaches with condyle resection have the risk of condyle instability, they have the advantage of removing the tumor in the hypoglossal canal and decompressing the hypoglossal nerve.

Anatomical considerations and transcondylar approach

Until now, the approach to the hypoglossal canal in hypoglossal canal meningioma has been the transcondylar approach, which has two directions. One is the lateral approach, and the other is the posterior approach. The lat-

Table 1 Reported cases of hypoglossal canal meningioma to date

a

Author, year	Side	Age	Sex	Symptom	Neurological deficit	Size (mm)	Cisternal portion (mm)	Extracranial portion (mm)	Tumor localization	Classification
Calzada, 2007[3]	Lt	62	F	Disequilibrium	Tongue atrophy	43×34	43×34	-	Cistern Hypoglossal canal	Cisternal type
Neeff, 2007[4]	Rt	28	M	Cervical mass Dysarthria	Hypoglossal nerve palsy	70×65	17×8	70×65	Cistern Hypoglossal canal Extracranial space	Dumbbell type
Zulkiflee, 2012[2]	Lt	54	M	Hoarseness Dysphagia	Tongue atrophy Shift of tongue Vocal fold paralysis	35×20	-	35×20	Hypoglossal canal Extracranial space	Extracranial type
Dobrowolski, 2016[5]	Rt	45	F	Headache	None	NA	NA	-	Cistern Hypoglossal canal	Cisternal type
Takahashi, 2019[6]	Rt	62	F	Snoring at night	Tongue atrophy Tongue decolorization, Shift of tongue	11×9	-	-	Hypoglossal canal	Canal type
Present case, 2021	Lt	65	F	Difficulty in speech and swallowing	Tongue atrophy Shift of tongue	9.4×6.3	9.4×6.3	-	Cistern Hypoglossal canal	Cisternal type

b

Author, year	Surgical approach	Condyle resection	Extent of resection	Hypoglossal nerve	Pathology	Postoperative course	Hypoglossal nerve palsy	Follow (month)
Calzada, 2007[3]	Far lateral app.	+	Subtotal removal	Preserved	NA	Pseudomeningocele No symptom of dysphagia	NA	48
Neeff, 2007[4]	Transcervical app.	-	Total removal (extracranial lesion) Not removed (intracranial lesion)	Sacrificed	Transitional meningioma	Gastrostomy feeding Dysarthria improved	impaired	2
Zulkiflee, 2012[2]	Transcervical app.	-	Total removal	NA	NA	Uneventful	NA	48
Dobrowolski, 2016[5]	Midline subtonsillar app.	-	Total removal	Sacrificed	Psammomatous meningioma	NA	NA	73
Takahashi, 2019[6]	Transcondylar app.	+	Total removal	Preserved	Transitional meningioma	Improved	Improved	14
Present case, 2021	Midline suboccipital app.	+	Total removal	Preserved	Meningothelial meningioma	Improved	Improved	9

eral approach is optimal if the goal is to remove only the tumor in the hypoglossal canal. The advantage of this approach is that it allows for safe removal without intracranial exposure. In contrast, the posterior approach is appropriate for the primary removal of tumors from the intracranial space within the hypoglossal canal. The most important point to note when using both approaches is the direction of the hypoglossal canal. The hypoglossal canal exists at a 45-degree angle in the 2 o'clock and 10 o'clock directions.[7] Therefore, the lateral approach reaches the hypoglossal canal almost perpendicularly, whereas the posterior approach reaches the hypoglossal canal at a 45-degree angle. Therefore, bone drilling should be performed with attention to the difference in depth between the medial and lateral sides of the hypoglossal canal in the posterior approach.[7] The use of navigation may solve this problem.

Surgical tips for hypoglossal canal meningioma

The hypoglossal venous plexus and instability of the craniovertebral junction (CVJ) require attention in hypoglossal

canal meningiomas. The venous plexus in the hypoglossal canal (anterior condylar vein) surrounds the hypoglossal nerve for its entire length in the hypoglossal canal.[7] With small tumors, the venous plexus is not occluded by the tumor. The operation would be difficult because of bleeding from the venous plexus. In our case, the tumor was small, and preoperative imaging studies showed that the venous plexus in the hypoglossal canal was expected to be open. Therefore, we were prepared intraoperatively and were able to manage bleeding from the venous plexus. Takahashi et al. emphasized the stability of the (CVJ) after drilling the condyle using a transcondylar approach. In previous anatomical studies, drilling up to 50% of the condyle was acceptable from the viewpoint of CVJ stability.[2]

Hypoglossal nerve preservation

Regarding the preservation of the hypoglossal nerve, Takahashi et al. described that it is difficult to preserve the function of sensory cranial nerves compressed by meningiomas even after total tumor removal; however, there is a possibility of preserving motor cranial nerves.[2] Cranial nerve monitoring is useful for preserving nerves, and reportedly for the hypoglossal nerve.[8] In our case, there was preoperative hypoglossal nerve palsy. Intraoperative hypoglossal nerve monitoring detected no waveform. However, improvement of the hypoglossal nerve palsy was observed immediately after surgery. Thus, even if there is no signal from the hypoglossal nerve on the monitor, there is a chance for symptomatic improvement if the nerve is morphologically preserved. However, more cases are needed to verify this finding.

Radiotherapy

There is no literature on radiation therapy for meningiomas arising from the hypoglossal canal. Radiotherapy is less invasive for asymptomatic lesions because it does not require a craniotomy.[9] However, in patients with nerve compression symptoms caused by tumors, decompression of the nerve is the goal of treatment; therefore, removing the tumor by craniotomy is necessary instead of radiation therapy.

Conclusion

We report a case of meningioma arising from the hypoglossal canal and review the literature on hypoglossal canal meningiomas. During surgical treatment, it is important to choose the appropriate approach depending on tumor localization and surgical purpose, and to be familiar with hypoglossal canal anatomy.

Previous Presentation

The manuscript was accepted as a podium presentation at the 33rd annual meeting of the Japanese Society for Skull Base Surgery on July 2-3, 2021, in Tokyo, Japan.

Supplementary Material

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Informed Consent

Informed consent was obtained from the patient for publication of this case report.

Conflicts of Interest Disclosure

The authors have no conflicts of interest regarding this paper.

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