

Spatial Relationship between Hypoglossal Schwannoma and the Vertebral Artery Using the Far-Lateral Approach

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Abstract

In hypoglossal schwannoma removal via the far-lateral approach needs care as the vertebral arteries are usually adjacent to the tumors. Thus, it is important to understand their location respective to schwannoma to conduct a safe surgery. We reviewed the data of eight patients with hypoglossal schwannoma who underwent surgery in Keio University Hospital in 2005–2013. There were five males and three females (mean age at initial presentation was 48.6 years, range 38–72 years). We especially focused on the spatial relationship between the vertebral artery and the tumor, and evaluated their spatial relationship from intraoperative findings. All eight hypoglossal schwannomas included in the current study were type B according to Kaye's classification. As for spatial relationship between the tumor and the vertebral artery, in six out of eight cases, the vertebral artery was located inside or beneath the tumor; in contrast, in the other two cases, it was pushed out by the tumor and identified just after dural opening. Through the far-lateral approach, we found that the vertebral artery was located inside or beneath in most hypoglossal schwannoma; however, the vertebral artery was occasionally located on the tumor surface. From an anatomical perspective, we speculate this unique location of the vertebral artery in these cases is due to the unusual course of the hypoglossal nerve of tumor origin.

Key words: hypoglossal nerve, vertebral artery, hypoglossal schwannoma, far-lateral approach

Introduction

Hypoglossal schwannoma are rare tumors; only approximately 100 cases have been reported to date.¹⁾ Most of the previous reports on this clinical entity were single case reports, and as far as we review, only a single case series was reported by Nonaka et al., in which the authors reported the usefulness of the extreme lateral infrajugular transcondylar-transtubercular exposure approach on 13 patients with hypoglossal schwannomas.²⁾

The origin of this tumor is the hypoglossal nerve, which arises along the front of the inferior olive as a series of rootlets that pass posteriorly to the vertebral artery as they converge on the dural orifice of the hypoglossal canal.³⁾

The far-lateral approach is useful for hypoglossal schwannoma removal. Rhoton divided the approach into three anatomic stages: muscular dissection, extradural dissection, and intradural exposure.⁴⁾ In the intradural exposure stage, the vertebral artery and its branches, the lower and upper cervical nerves, are important anatomical figures to which special

attention should be paid during surgery.^{3,4)} These anatomical structures are usually deformed by tumors in patients with hypoglossal schwannoma, and such tumors should be carefully removed without damage to the normal vascular and neuronal structures.

Recently, we experienced a case of hypoglossal schwannoma in that the vertebral artery was found just after dural opening atop of the tumor. It was very unusual from our experience. In most cases with hypoglossal schwannomas, the vertebral artery passed either through or beneath the tumor via the far-lateral or suboccipital approach. In the present study, we performed anatomical consideration of hypoglossal schwannoma and placed special focus on the spatial relationship between the tumor and vertebral artery in eight patients with hypoglossal schwannoma who underwent surgery at Keio University Hospital over the past 9 years.

Patient Characteristics

Eight patients with hypoglossal schwannoma underwent surgical tumor removal at Keio University

Table 1 Clinical and radiological presentation of schwannomas in the current series

Case	Age Sex	Kaye's type	Symptoms	Surgical approach	T ₂	Gd* ¹	Cyst* ²
1	39M	B	XII	Midline suboccipital	Iso-high	+	-
2	57F	B	XII, P	Rt. transcondylar	High	-	+
3	39M	B	X, XII, P, dysarthria	Lt. transcondylar	Iso	+	-
4	72F	B	XII, A, Rt. hemiparesis	Lt. lateral suboccipital	Iso-high	+	+
5	56F	B	X, XII, P, A	Lt. transcondylar	Iso	+	+
6	43M	B	IX, X, XII, A	Rt. transcondylar	Iso-high	+	+
7	45M	B	VI, X, XII, P	Lt. transcondylar	High	+	+
8	38M	B	XII	Lt. transcondylar	Iso-high	+	+

A: ataxia, F: female, Lt.: left, M: male, P: nuchal pain, Rt.: right, Type A: intracranial tumor, Type B: intra- and extracranial tumor, Type C: extracranial tumor (Kaye, 1984)⁵⁾, *¹: gadolinium enhancement of the tumor in T₁-weighted magnetic resonance (MR) image, *²: cyst formation of the tumor in T₁-weighted MR image.

Hospital in 2005–2013. Written informed consent was obtained from all patients included in this study and was approved by the Institutional Review Board at Keio University. All patients consented to a complete neurological examination as well as any required radiological examinations at the time of admission.

Methods

We have reviewed the clinical charts as well as operative video of patients with hypoglossal schwannoma retrospectively. Especially, we assessed the spatial relationship between the vertebral artery and the tumor from intraoperative findings.

Results

I. Clinical manifestations

All the patients in this study suffered from 12th nerve palsy. Vagal nerve palsy was the second frequently observed symptom. Nuchal pain was also observed in three patients (Table 1).

II. Radiological findings

All eight hypoglossal schwannomas were type B according to Kaye's classification.⁵⁾ All the tumor was high-intensity in T₂-weighted magnetic resonance (MR) image, and all but one schwannoma was enhanced by gadolinium in T₁-weighted MR image. Cyst formation was observed in six out of eight patients included in the current study (Table 1).

III. Surgical approaches

The far-lateral transcondylar approach was adopted in six cases, midline suboccipital approach in one case, and lateral suboccipital approach in one case (Table 1).

Table 2 Intraoperative findings of schwannomas in the current series

Case	Location of vertebral artery in relation with tumor	Origin of the tumor
1	VA located beneath the tumor	XII
2	VA located beneath the tumor	XII
3	VA located beneath the tumor	XII
4	VA located on the surface of the tumor	XII
5	VA located on the surface of the tumor	C1 root of XII
6	VA located beneath the tumor	XII
7	VA located beneath the tumor	XII
8	VA located beneath the tumor	XII

VA: vertebral artery.

IV. Spatial relationship between the vertebral artery and the tumor

The intraoperative findings of the eight cases were assessed and are summarized in Table 2. In six cases, the vertebral artery passed either through or beneath the tumor via the far-lateral or suboccipital approach. However, in the other two cases, the vertebral artery was pushed out by the tumor and identified just after dural opening.

Illustrative Cases

I. Case 5

A 56-year-old female referred to our hospital with the chief complaint of left 10th and 12th nerve palsy, ataxia, and nuchal pain. Preoperative MR images of the patient are shown in Fig. 1(A–E). This

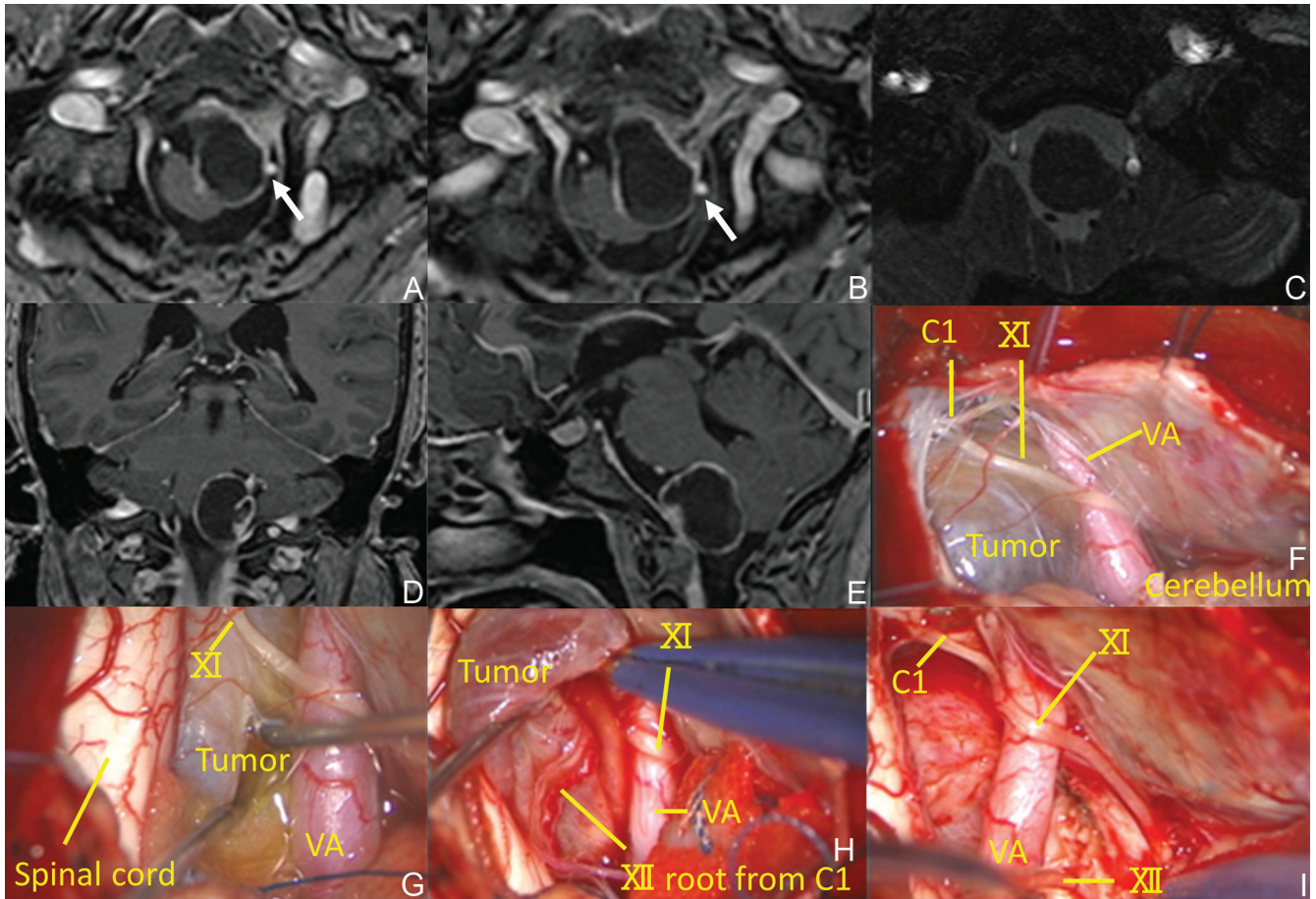


Fig. 1 A, B, D, E: T₁-weighted magnetic resonance images with gadolinium enhancement of case 5 (A and B: axial; D: coronal; E: sagittal). C: Original time of flight magnetic resonance image of case 5 shows spatial relationship between right vertebral artery and hypoglossal nerve. F–I: Intraoperative images of case 5. In this case, the surgery was performed via the left transcondylar approach. The vertebral artery was located on the surface of the tumor and identified just after the dural opening.

patient underwent surgery via the left far-lateral transcondylar approach with a preoperative diagnosis of hypoglossal schwannoma. In this case, the vertebral artery was identified just after dural opening (Fig. 1F) that was different than usual. Opening the cystic part of the tumor (Fig. 1G), detachment of the arachnoid and dentate ligaments revealed that the C1 root nerve located anteriorly to the vertebral artery posteriorly to the tumor, and buried within the tumor. This C1 root nerve joined the cranial root of the hypoglossal nerve near the dural orifice of the hypoglossal canal and ran anteriorly to the vertebral artery (Fig. 1H). Pathological examination revealed that the tumor specimen was a schwannoma. Since it passed the hypoglossal canal (forming a dumbbell-shaped tumor) and originated from the C1 rootlet, which connects to the hypoglossal nerve near the hypoglossal canal, we determined that it originated from the C1 rootlet of the hypoglossal nerve. Time of flight (TOF) original data (Fig. 1C)

showed that contralateral hypoglossal nerve run posteriorly to the vertebral artery as usual.

II. Case 6

Six of eight cases had quite parallel surgical findings. This patient (Case 6) is a 43-year-old male with hypoglossal schwannoma clinically revealed as 9th, 10th, right 12th palsy, and ataxia. MR images of the patient are shown in Fig. 2(A–C). The patient underwent surgery via the right far-lateral transcondylar approach. Surgical findings of this case showed that the vertebral artery passed beneath the tumor in the operative field, and the vertebral artery was identified only after tumor removal (Fig. 2D–F).

Discussion

In the present study, in six out of eight cases of hypoglossal schwannoma, the vertebral artery passed either through or beneath the tumor via

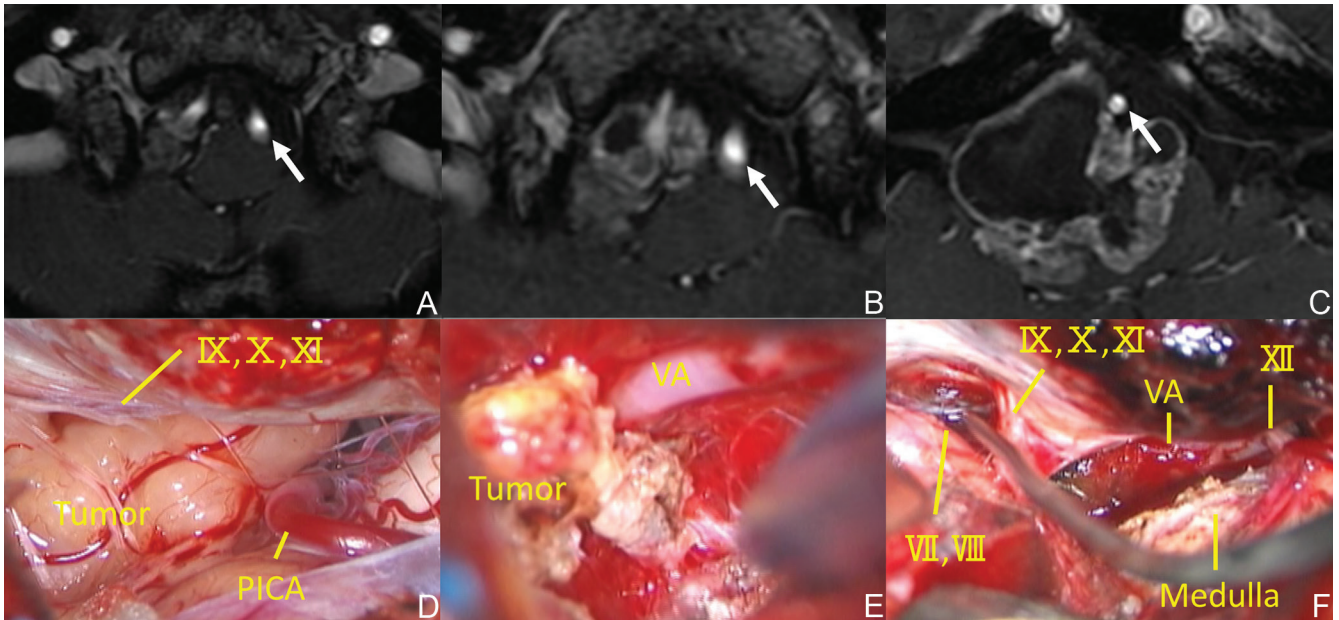


Fig. 2 A–C: Axial T₁-weighted magnetic resonance images with gadolinium enhancement of case 6. D–F: Intraoperative images of case 6. In this case, the surgery was performed via the right transcondylar approach. The vertebral artery was located beneath the tumor and identified only after tumor relocation.

the far-lateral or suboccipital approach, while the vertebral artery was located on the surface of the tumor in two cases.

In normal anatomy, the hypoglossal nerve arises along the front of the inferior olive as a series of rootlets that pass posteriorly to the vertebral artery and picks up a branch from the anterior ramus of C1 as they converge on the dural orifice of the hypoglossal canal.³⁾ Accordingly, the vertebral artery is pushed by the tumor anteriorly, and normally located beneath the tumor via transcondylar approaches. According to this, Sato et al. mentioned that hypoglossal schwannoma usually compressed the medulla and the junction of the vertebral artery and posterior inferior cerebellar artery medially.⁶⁾ However, in cases 4 and 5 of the current series, the vertebral artery was found atop the tumor just after dural opening. Of note, the clinical symptoms did not differ among all cases, suggesting that the interpretation of preoperative MR images is important for differentiating cases in which the vertebral artery is located beneath the tumor.

Case 5 of the current series is the first reported case of a hypoglossal schwannoma originating from the C1 rootlet of the hypoglossal nerve. In this case, one of the rootlets of the hypoglossal nerve ran inferiorly toward the spinal canal and joined the C1 rootlet. The intraoperative findings suggested that this rootlet of the hypoglossal nerve joining the C1 rootlet was the origin of this schwannoma.

We speculate in case 5 that hypoglossal schwannoma originally arose from this junction of the hypoglossal nerve and the C1 rootlet and developed in the extracranial direction via the hypoglossal canal. Normally, hypoglossal nerve run posteriorly to the vertebral artery toward hypoglossal canal, however, in this case, tumor originated from hypoglossal nerve run anteriorly to the vertebral artery, and the tumor in this case pushed out the vertebral artery posteriorly in consequence, and resulted in this unique spatial relationship between the vertebral artery and the tumor. It was reported that the cerebellar arteries appear in the late branchial phase in a plexiform pattern lying between the facial and hypoglossal nerve roots, and the plexiform configuration can give rise to wide variations in the origin and course of the branches of the basilar and vertebral arteries by the late retention of different remnants of the plexiform vessels.⁷⁾ Thus, it is not considered to be very unusual to find variations in the relationship between hypoglossal nerve root and vertebral artery.

In summary, we speculate if hypoglossal schwannoma originates from hypoglossal nerve that passes posteriorly to the vertebral artery (indicated by blue circle in Fig. 3; normal anatomy), the vertebral artery passed either through or beneath the tumor via the far-lateral or suboccipital approach. On the other hand, if hypoglossal schwannoma originates from hypoglossal nerve that passes anteriorly to

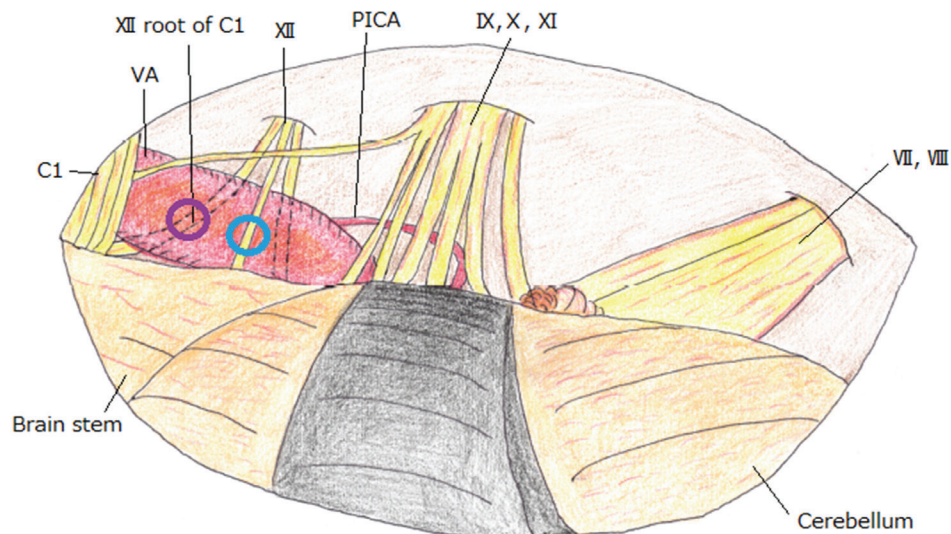


Fig. 3 Anatomical relationship between the tumor and the vertebral artery. If the tumor origin rootlet of the hypoglossal nerve ran caudally to the vertebral artery as in normal anatomy, then the hypoglossal neurinoma originated from the point indicated by the *blue circle*. If the tumor origin rootlet of the hypoglossal nerve ran rostral to the vertebral artery, the hypoglossal neurinoma originated from the point indicated by the *purple circle* and pushed out the vertebral artery.

the vertebral artery (indicated by purple circle in Fig. 3; normal variant), the vertebral artery is pushed out by the tumor and found atop the tumor via the far-lateral or suboccipital approach.

Our small study population is this study's main limitation. However, since only about 100 other cases have been reported to date, the accumulation of further cases will come in time.

In conclusion, in cases of hypoglossal schwannoma, the vertebral artery usually passed either through or beneath the tumor via the far-lateral or suboccipital approach. In some cases, the vertebral artery was identified just after the dural opening.

From an anatomical perspective, we speculate that this unique location of the vertebral artery in the case is due to the unusual course of the hypoglossal nerve.

Conflicts of Interest Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this article.

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