Original Article

Vertebral artery is an anatomical landmark in the posterior unilateral resection of cervical benign nerve sheath tumors with dumbbell extension of Eden type 2 or 3

ABSTRACT

Background: In the cervical nerve sheath tumor (NST) surgery with dumbbell extension of Eden type 2 or 3, selection of anterior, posterior, or combined approach remains controversial.

Objectives: This technical note aimed to propose possible advantages of the posterior unilateral approach (PUA). Methods: Six patients who underwent the surgical treatment of cervical NSTs with dumbbell extension of Eden type 2 or 3 were included. The critical surgical steps included (1) complete separation of extradural and intradural procedures, (2) careful peeling of the neural membranes (epineurium and perineurium) from the tumor surface in the extradural procedure, (3) complete removal of the extradural tumor within the neural membranes, (4) intradural disconnection of tumor origin, and (5) intentional tumor removal up to the vertebral artery (VA), i.e., the VA line.

Results: The tumor location of dumbbell extension was Eden types 2 and 3 in two and four patients. Gross total resection was achieved in two patients and intentional posterior removal of the tumor to the VA line was achieved in the remaining four patients. No vascular or neural injuries associated with surgical procedures occurred. Postoperative neurological assessment revealed no symptomatic aggravation in all patients. No secondary surgery was performed during the study period.

Conclusion: PUA was safe and less invasive for functional recovery and tumor resection, if the anatomical relationship between the tumor and VA is clearly understood. The VA line is an important anatomical landmark to limit the extent of tumor resection.

Keywords: Cervical, dumbbell tumor, nerve sheath tumors, schwannoma, vertebral artery

INTRODUCTION

In cervical nerve sheath tumor (NST) surgery with dumbbell extension of Eden type 2 or 3, several surgical issues are encountered, such as preservation or sacrifice of root fibers critical for upper extremity motor function (C5–C8), dealing with the vertebral artery (VA) and surrounding venous plexus and preservation or reconstruction of spinal stability.[1,2] Although the anterior and posterior combined approaches may be necessary for large tumors extending inside and outside the cervical canal, its indication is generally limited.[3-7] The surgical approach for small-to-moderate tumors remains controversial. Whether complete resection at the initial surgery for such benign NSTs is necessary remains unclear. On this technical note, we proposed the technical advantage of the posterior unilateral approach (PUA) for cervical benign NSTs with dumbbell extension of Eden type 2 or 3. The VA

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line was proposed as an anatomical landmark formed by VA and the outer edge of the neural foramen, where the spinal sensory ganglion is located at the C3–C6 level.

PROCEDURE

Subjects

Patients with cervical NSTs with dumbbell extension of Eden type 2 or 3 who underwent surgical treatment at our institution from October 2020 to May 2022 were included in this study. The study patients consisted of two males and four females, with an average age of 47 years. All patients underwent comprehensive evaluation both pre and postoperatively. The initial symptoms were mild in all patients, such as cervical pain or paresthesia of unilateral upper extremity. The minimum duration of postoperative follow-up was 6 months. Medical charts of all patients were retrospectively reviewed for the spinal level, tumor location, surgical findings, pathological diagnosis, and surgery-related outcome. The tumor location of dumbbell extension was classified by intraoperative inspection based on the Eden classification.

Surgical technique

All surgical procedures were conducted by neurophysiologically monitoring motor-evoked potential and sensory-evoked potential. In patients with cervical NSTs originating from C1 or C2 nerve roots, PUA with or without lateral intermuscular approach on the tumor side was applied. In patients with cervical subaxial NSTs, PUA with minimal resection of the ipsilateral facet joint was designed to prevent postoperative instability of the cervical spine. The distal end of tumor resection was limited to the VA line, an anatomical landmark formed by the outer edge of the neural foramen at the C3–C6 level, where the spinal sensory ganglion is located. Critical surgical steps included (1) complete separation of extradural and intradural procedures to prevent postoperative cerebrospinal fluid leakage, (2) careful peeling of the neural membranes (epineurium and perineurium) from the tumor surface in the extradural procedure [Figure 1a], (3) completely tumor removal of extradural procedure within the neural membranes, (4) intradural inspection and disconnection of the tumor origin of anterior or posterior nerve root fibers [Figure 1b], and (5) intentional tumor removal up to the VA line, formed by VA and the outer edge of the neural foramen [Figure 1c and d].

Postoperative assessment

The spinal level of the tumors was classified based on the spinal nerve root level of the tumor origin; C1 in 1, C2 in 1, C3 in 1, C4 in 1, and C5 in 2 patients. The tumor location of dumbbell extension was Eden types 2 and 3 in two and four patients, respectively. An association with neurofibromatosis was suggested in one of six patients. Gross total resection was achieved in two patients and intentional posterior removal of the tumor limited to the VA line was achieved in the remaining four patients. The operative time was from 319 to 388 (mean, 354) min, and the intraoperative blood loss ranged 30–70 (mean, 47) ml. No vascular or neural injuries associated with surgical procedures. Pathological diagnosis confirmed the benign schwannoma in all patients. Postoperative neurological assessment revealed transient sensory deficits in three patients but completely resolved by 3 months postoperatively. Postoperative outpatient neurological assessment demonstrated no symptomatic aggravation of neurological function postoperatively in all patients. No secondary surgery to remove the remaining tumor was performed during the study period. Patient characteristics are summarized in Table 1.

Illustrative case: Case 3

A 42-year-old woman presented with discomfort in the left upper extremity. Magnetic resonance imaging (MRI) revealed cervical extramedullary tumor with dumbbell extension at the C2/C3 level on the left side [Figure 2a]. The tumor demonstrated homogeneous enhancement on T1-weighted contrast MRI and was localized in the spinal canal and extended distal to the intervertebral foramen. Left PUA was applied at the C2/C3 level. Unilateral partial facetectomy on the tumor side was designed to involve <25% of the facet joint to prevent postoperative instability of the cervical spine. Intradural inspection demonstrated that the tumor originated from C3 posterior roots. Intentional posterior removal

Table 1: Characteristics of patients with cervical nerve sheath tumors with dumbbell extension of Eden type 2 or 3

Patient number	Age	Sex	Spinal level	Association of NF	Removal of the tumor	Operation time	EBL (mL)	Eden classification	Pathology	Postoperative neurological complications	Postoperative follow-up (months)
1	56	Male	C4	–	IPR-VA	6:28	55	2	Schwannoma	-	24
2	65	Female	C2	–	Total	6:05	70	2	Schwannoma	-	20
3	42	Female	C3	–	IPR-VA	5:51	40	3	Schwannoma	-	19
4	48	Female	C1	–	Total	5:19	30	3	Neurofibroma	-	16
5	47	Female	C5	–	IPR-VA	5:35	50	3	Schwannoma	-	7
6	24	Male	C5	+	IPR-VA	6:05	35	3	Neurofibroma	-	6

IPR-VA - Intentional posterior removal of the tumor up to the vertebral artery line; NF - Neurofibromatosis; EBL - Estimated blood loss

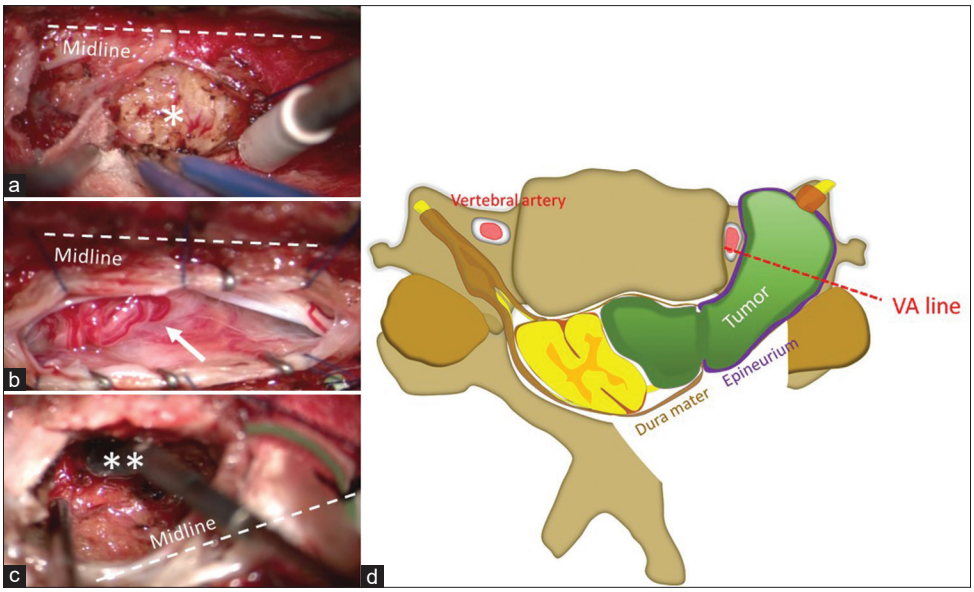

Figure 1: Intraoperative images showing the surgical steps in case 3 (C3 level, Eden type 3) (a-c) and the schematic image showing the concept of vertebral artery line (d). (a) The neural membranes (epineurium and perineurium) were incised longitudinally to the nerve root to expose the tumor surface (operative view from the ipsilateral side of tumor). Please note the midline (----) and tumor surface (*), (b) Intradural inspection revealed that tumor originated from left C3 posterior roots (arrow), (c) Tumor removal was limited just behind the vertebral artery that was located beyond the epineurium and confirmed by using vascular doppler (**) (Operative view from the opposite side of tumor)

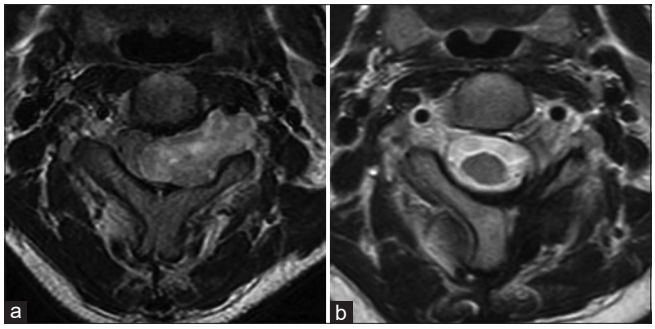

Figure 2: Pre- and post-operative T2-weighted axial magnetic resonance imaging (MRI) of case 3. (a) Preoperative MRI shows cervical extramedullary tumor with dumbbell extension at the C2/C3 level on the left side, (b) Postoperative MRI shows the intentional posterior removal of the tumor to the vertebral artery line

of the tumor limited to the VA line was achieved without any vascular or neural complications. The tumor location of dumbbell extension was Eden type 3. Postoperative MRI revealed that the intentional posterior removal of the tumor to the VA line was successfully achieved [Figure 2b]. Neurological assessment at the recent outpatient clinic revealed the excellent condition of neurological function.

DISCUSSION

Spinal cord tumors with dumbbell extension account for approximately 15%–18% of all spinal cord tumors, which more likely occur in the cervical spine than in the thoracic or lumbar spine, and most of them are benign NSTs.[5,8] However, because the NST with dumbbell extension is frequently located at the intradural and extradural parts, postoperative cerebrospinal fluid leakage is a problem. It can be solved by separating intradural and extradural procedures.[9-11] Furthermore, to prevent damage to surrounding normal structures, such as the nerve roots, arteries, and venous plexus, tumor removal at the extradural section should be performed only within the epineurium.[9,10] The most critical point may be understanding the anatomical relationship between the tumor and VA. The VA course is generally constant with only anatomical minor variations.[12-14] Between the VA origin from the subclavian artery and the basilar artery junction, the VA can be divided into four segments: first, between the subclavian artery and transverse foramen of C6; second, within the transverse foramens of C6–C2; third, the tortuous segment (between the transverse foramens of C2–C1); and fourth, between the C1 and the basilar artery junction. The VA on the left side is generally larger than that on the right side. VA branches from the subclavian artery and mostly ascends into the transverse foramen of C6. The sympathetic chain extends anteriorly to the VA. The second segment of the VA ascends almost vertically through the transverse foramens of C6–C2. Within the transverse foramen, the VA crosses in front of the spinal nerve roots, where the spinal sensory ganglion is located. The VA is accompanied by vertebral veins, forming the vertebral venous plexus wrapping the VA. After leaving the transverse foramen of the C2, the VA forms a curve convex laterally to reach the transverse foramen of C1 and runs backward and curves upward the lateral mass of C1 to enter the VA groove of C1. After leaving the VA groove of C1, the VA runs upward

and forward and enters the foramen magnum as its anterior margin, perforating the dura mater over the C1 nerve root. The third segment of the VA is located within the suboccipital triangle limited by three muscles: the rectus capitis posterior major muscle on the medial side, the superior oblique muscle on the superolateral side, and the inferior oblique muscle on the inferolateral sides.[13] Hence, surgeons should understand the anatomical relationship between the tumor and VA in detail on preoperative images.

Although the anterior and posterior combined approaches may be necessary for large tumors extending inside and outside the cervical canal, the surgical approach for small or moderate tumors of Eden type 2 or 3 of cervical spine remains controversial [Table 2].[3-7,15-20] The anterior approach is one of the choices even when the tumor extends into the dura mater.[15,17] The anterior approach requires careful handling of VA because the tumor is located just behind the VA. Furthermore, to prevent postoperative deformity of the spinal column, spinal stabilization is frequently necessary for the anterior approach.[5,17] Conversely, the posterior approach requires facet joint resection to entirely expose the tumor that extends significantly beyond the intervertebral foramen. When aiming for total resection using a posterior approach alone, surgeons should resect a large part of the unilateral facet joint, resulting in the risk of postoperative instability of the cervical spine.[4,6,16,18,19] To preserve the postoperative stability of the spinal structure, tumor resection was performed using a posterior unilateral laminectomy without removing >25% of the facet.[20] In our surgical strategy, the lateral intermuscular approach can be added, or the tumor beyond the VA line can be left behind intentionally. The remaining tumor outside the VA line can be safely removed at the second-stage anterior surgery, especially using the retrojugular approach, as necessary. The surgical strategy presented here can be safely indicated for cervical benign NSTs with dumbbell extension of Eden type 2 or 3. Postoperative complications were minimal. The extraforaminal tumor outside the VA line requires careful follow-up from a medium-to long-term perspective, despite a favorable short-term outcome.

CONCLUSION

In cervical benign NST surgery with dumbbell extension of Eden type 2 or 3, how to protect VA is critical associated with surgical procedures. The PUA was safe and less invasive for functional recovery and tumor resection, if the anatomical relationship between the tumor and VA is clearly understood. The VA line is an important anatomical landmark to minimize the vascular or neural injuries and limit the extent of tumor resection.

Table 2: Literature summary of surgical approaches for cervical nerve sheath tumors with dumbbell extension

Authors (year)	Number of cases	Approach	Note
Hakuba *et al*. (1984)[15]	5	Anterolateral	Any cervical dumbbell-shaped tumor below the C2 level can be removed via an anterolateral approach as long as no more than three levels are involved
McCormick (1996)[18]	12	Posterior	Single-stage modified posterior midline exposure with laminectomy and complete unilateral facetectomy
Iwasaki *et al*. (1999)[17]	4	Anterior	Single-stage anterior approach with VA identification and corpectomy
Asazuma *et al*. (2004)[3]	35 7	Posterior Combined	Systematic, imaging-based three-dimensional characterization of the shape and location of cervical dumbbell tumors is essential for planning optimal surgery
Jiang *et al*. (2009)[5]	30 7 6	Posterior Anterior Combined	The recurrence rate significantly decreases after radial tumor resection. Revision surgeries are associated with more complications
Zhao and Xu (2009)[7]	12 4	Posterior Combined	Extensive posterolateral exposure enables surgeons to reach the lateral-most portion of cervical spine dumbbell tumors and facilitates VA septation and resection of vertebral body encroachment of the tumor
Tomii *et al*. (2013)[20]	5 13 1	Anterior Posterior Combined	When using a posterior approach, recapping laminoplasty using an ultrasonic bone curette is very useful to remove tumors without sacrificing facet joints
Gu *et al*. (2015)[4]	35 6	Posterior Combined	Total removal of intra- and extra-foraminal cervical subaxial schwannomas could be possible using a posterior approach with facet removal if the extraforaminal tumor size was <5.4 mm
Huang *et al*. (2017)[16]	26	Posterior	The one-stage posterior approach was feasible and effective for the treatment of dumbbell schwannomas in the subaxial cervical spine. The tumor could be completely removed in most cases safely
Ryu *et al*. (2019)[6]	62 5 5	Posterior Anterolateral Combined	Only high cervical level (OR: 5.48, $P=0.033$) and encased VA (OR: 0.07, $P=0.014$) were significant predictors for GTR
Singh *et al*. (2020)[19]	3 1	Posterolateral Combined	The posterolateral approach for cervical dumbbell giant neurofibromas is safe and effective and promises maximum safe excision

GTR - Gross total resection; VA - Vertebral artery; OR - Odds ratio

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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