PEER REVIEWED LETTER

Anterior Corpectomy Approach for Removal of a Cervical Intradural Schwannoma

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Spinal schwannomas are typically intradural-extramedullary neoplasms thought to arise from Schwann cells or their progenitors which occur proportionally throughout the spinal canal.[1-3] They most typically arise from dorsal sensory rootlets and occupy a posterior-lateral location in the spinal canal. Thus, posterior surgical procedures have become the conventional method to remove these tumors providing adequate exposure in most cases. More anteriorly located tumors may be approached through a posterolateral direction with section of the dentate ligament and gentle rotation of the spinal cord.[1,3,4] However, posterior and posterolateral approaches may be problematic for removing tumors located in the midline and ventral to the spinal cord.

Although the anterior approach has been applied widely to treat cervical spondylosis, it has rarely been used to remove intradural tumors. [5] Here, we present a case of a ventral cervical spinal schwannoma removed through an anterior approach followed by spinal reconstruction.

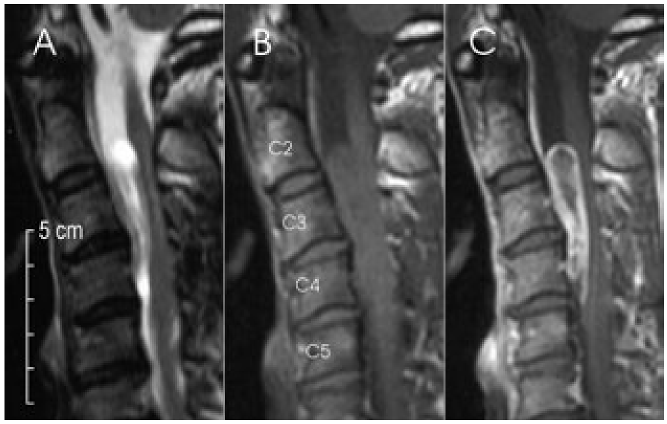

Figure 1. *Sagittal magnetic resonance images from C2-C6 prior to surgery. (A) T2 weighted images showed an intradural extramedullary tumor with cystic components located behind the C2/3 disc and extending to behind the C4 vertebral body. (B) T1 sequences showed the tumor to be isointense with the spinal cord. (C) Contrast enhancement was present throughout the tumor but especially around the capsule.*

Case Report

History and presentation. A 31-year-old man with no other medical concerns presented with a ten month history of neck pain. Pain radiated to the retro-auricular region on the right side and was exacerbated by coughing and straining. He also complained of progressive gait difficulties and bilateral arm weakness and clumsiness.

Examination. On physical examination, cranial nerve function was normal. Motor testing exhibited grade 4/5 power in deltoids, biceps and triceps bilaterally. The remaining upper extremity motor exam and the lower extremity exam were normal. Sensory examination revealed decreased sensory perception to pain and temperature between C5 and T1 bilaterally, right more prominent than left. This deficit was most pronounced in the C5 dermatome. Reflexes were brisk and symmetric with spreading reflexes at the brachial radialus bilaterally and at the right biceps. Toes were down-going however there was ankle clonus on the right side. Gait appeared normal although the patient had difficulty with both plantar and dorsi-flexion when carrying his weight.

Radiology. A cervical spine magnetic resonance imaging scan revealed an intradural-extramedullary tumor located anteriorly in the spinal canal. The tumor extended from the lower aspect of C2 to the inferior endplate of the C4 vertebral body (Figure 1). In the transverse plane the tumor occupied a midline anterior position with direct posterior displacement of the spinal cord (Figure 2).

Operative Procedure. Owing to the direct posterior displacement of the spinal cord the posterior surgical approach to the tumor was felt to be unfavorable. Tumor resection was therefore undertaken through an anterior approach. The patient was positioned supine with his head rotated towards the left side and in slight extension. A standard approach to the anterior cervical spine through a longitudinal incision along the anterior border of the sternocleidomastoid was undertaken. Longus colli insertions between C2 and C5 were dissected and self-retraining retractors were applied. Distraction was applied between C2 and C5 using interosseous distraction pins. Discectomy was

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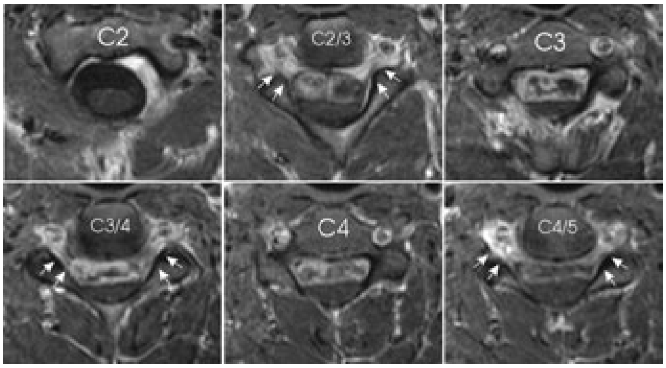

Figure 2. *Axial, contrast enhanced T1 weighted magnetic resonance images between C2 and C5 demonstrated an intradural extra-axial tumor in the anterior spinal canal. The lesion was located in the midline and the spinal cord was displaced posteriorly without any lateral displacement or rotation. Posterior access would require partial removal of the lateral masses bilaterally. Arrows indicate the C3, C4 and C5 nerve root foramena which were not involved or widened by the tumor.*

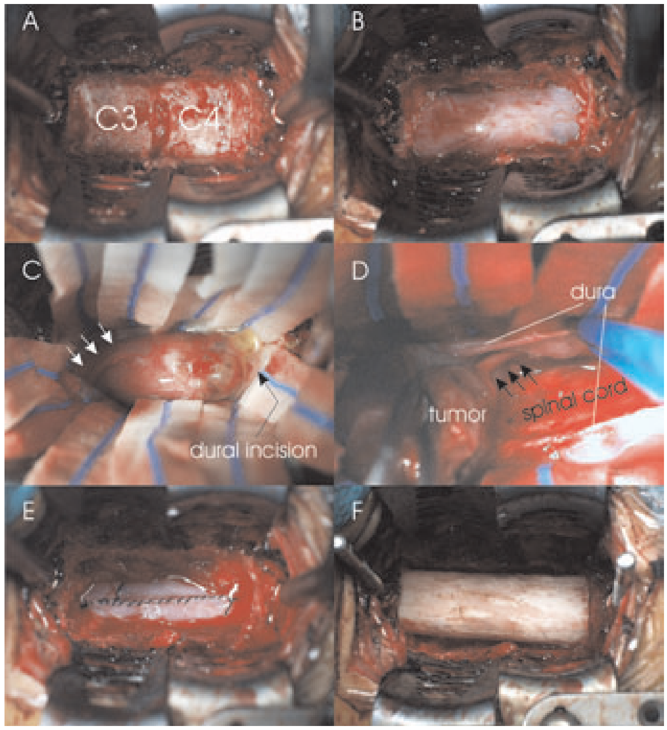

Figure 3. *Operative technique. A standard anterior approach to the upper subaxial spine was undertaken. (A) partial corpectomies of C3 and C4 with lateral resection of the uncovertibral joints on each side of the midline. (B) Completed corpectomies demonstrating dural exposure. The dura was incised in the midline exposing the tumor. (C) At the superior aspect of the tumor a fibrovascular band attached to the tumor from a rostral direction (arrows). (D) The tumor was dissected off the spinal cord and its attachment to the C4 nerve root (arrows) was ligated. (E) The dura was closed in a running water-tight fashion. (F) Fibular allograft was used to span the bony defect.*

performed at C2/C3, C3/C4, and C4/C5. A high-speed pneumatic drill was used to perform wide corpectomies of C3 and C4 (Figure 3). The posterior longitudinal ligament was removed and further bone was removed to the level of the pedicles maximizing dural exposure. Significant epidural bleeding was encountered from the epidural venous plexus which was controlled with microfibrillary collagen. The dura was opened in the midline over the entire extent of its exposure. The inferior aspect of the tumor was identified and was mobilized using both blunt and sharp micro-dissection. Several arachnoid adhesions to the spinal cord were identified and divided. The tumor was found to be contiguous with a portion of the C4 anterior nerve root. In addition there appeared to be a vascular bundle supplying the tumor from a rostral anterior location near the midline on the right side (Figure 3). Both these structures were taken to mobilize the tumor. The superior aspect of the tumor was gently delivered into the corpectomy defect through the dura allowing gross total resection. As the last portion of the tumor was removed decompression of the epidural venous plexus resulted in significant bleeding. This was difficult to control with microfibrillary collagen but responded to tamponade with strips of gelatin sponge. More than 10 liters of blood was lost during the hour required to achieve hemostasis after tumor removal. The dura was closed in a running fashion using nylon suture (Figure 3). Allograft fibula was used to span between the decorticated endplates of C2 and C5 and an Atlantis anterior cervical plate (Metronic Sofamor Danek, Inc., Memphis, TN) was applied with fixation to C2 and C5 as well as to the allograft (Figure 4). The wound was closed in layers. Estimated blood loss for the procedure was 13 liters.

Pathological Studies. Histological sections showed an encapsulated tumor with the features of a schwannoma. The

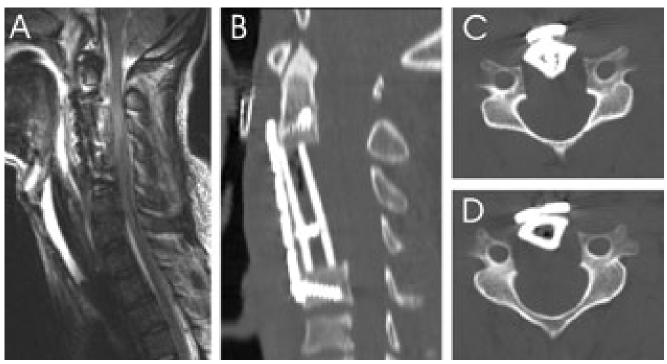

Figure 4. *(A) Postoperative magnetic resonance imaging demonstrated complete resection of the tumor and relief of spinal cord compression. (B) Postoperative CT scans demonstrated expansion of the spinal canal and with proper positioning of the instrumentation and allograft. Axial images demonstrated the wide exposure of the spinal canal achieved anteriorly through the corpectomies at C4 (C) and C3 (D) with resection to the level of the pedicles.*

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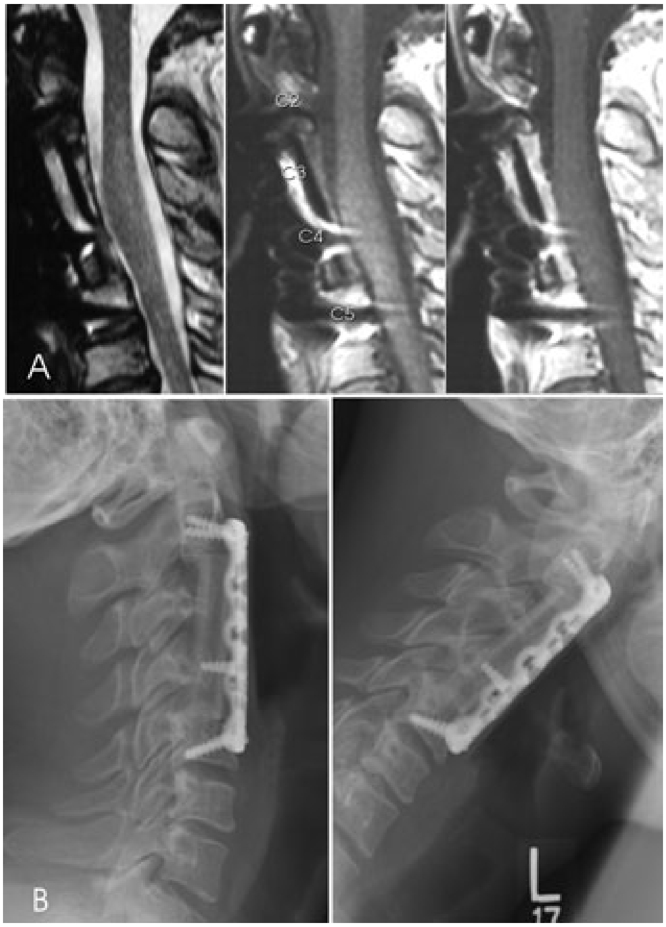

Figure 5. *(A) Magnetic resonance sequences six months after surgery. Panels from left to right are T2, T1 and contrast enhanced T1 images. The spinal cord had re-expanded, decompression was complete and there was no evidence of residual tumor. (B) Six month extension and flexion lateral radiographs showed improved lordosis compared to the pre-operative images and stability through the construct spanning C2 to C5.*

tumor exhibited pseudoglandular cystic degeneration. Many hyalinized vessels were intermingled with foci of iron and lipid containing macrophages. Proliferative activity was largely confined to the mononuclear inflammatory infiltrate. The tumor was immunopositive for S100. Neurofilament proteins immunohistochemistry was also used to further distinguish from neurofibroma.

Post-operative course. The patient's postoperative neurological exam was unchanged from his pre-operative state. He was immobilized in a hard collar and discharged from hospital uneventfully. He was free of pain. Cervical spine MRI and CT scan showed good decompression of the spinal cord and no evidence of residual tumor (Figure 4).

At the three and six month postoperative evaluations the patient remained pain free and had recovered full neurological function. By six months his neurological exam was normal. Static and dynamic cervical spine X-rays demonstrated a stable construct between C2 to C5 and the external orthosis was discontinued at three months. Repeat MRI at six months did not reveal any tumor recurrence (Figure 5).

Discussion

Spinal cord tumors account for 15% of central nervous system (CNS) neoplasms.[6] Approximately 25% of spinal cord tumors in adults are nerve sheath tumors.[2,4] Distinguished from neurofibromas, schwannomas are typically smooth masses with a discrete capsule and with a focal attachment to the involved nerve. In the spinal canal they are usually dissected easily from surrounding structures.[1,7] Prognoses with surgical excision is excellent and the life expectancy of these patients parallels that of the general population.[8,9] However, the rate of clinical recurrences after subtotal resection is 50%[3] with repeat operations carrying a high risk of neurological morbidity. Thus, full exposure with confident gross total resection is desirable at first operation.

Most spinal schwannomas arise from dorsal sensory nerve rootlets and are therefore located dorsolateral to the spinal cord.[3,8] However, approximately 23% of cervical nerve sheath tumors are located ventrally.[8,10] Most of these represent neurofibromas arising from ventral nerve rootlets. In addition, approximately 1% of schwannomas present as an intramedullary mass and are thought to arise from the perivascular nerve plexi, from neural crest progenitor cells displaced during embryogenesis, or by pial cell metaplasia.[11-13] Purely midline ventrally located intradural Schwannomas with direct posterior displacement of the spinal cord are quite rare.[5] Thirty percent of nerve sheath tumors extend through the dural root sleeve forming a dumbbell tumor with both an intradural and an extradural component. This frequency is elevated in the cervical spine (approximately 50%) likely because the intradural root segment is short.[8]

Traditionally, surgeons have approached all these tumors through a posterior approach. Indeed, the vast majority of intradural-extramedullary tumors can be well visualized and resected through a standard posterior laminectomy.[2,4] Most ventrally located intradural-extramedullary schwannomas are eccentric to the spinal cord and displace it laterally. These tumors may also be accessed through posterior laminectomy with the addition of facet and/or pedicle resection. If necessary additional ventral exposure may be achieved by sectioning the dentate ligament or non-critical dorsal nerve roots to allow gentle rotation of the spinal cord.[4] However, these modifications raise several concerns. Retraction or rotation of the spinal cord to expose the lesion may lead to spinal cord injury.[5,14] It is for principally this reason that many argue that ventral cervical and thoracic extradural lesions should be approached anteriorly rather than through a more conservative posterior approach. This argument however has generally not been applied when addressing intradural pathology. In addition, partial or complete unilateral or bilateral facetectomy may predispose the patient to postoperative instability of the cervical spine. The development of a "Swan-neck" deformity has been associated with extensive cervical laminectomy.[7,15] Instrumentation and arthrodesis of the contralateral intact facet joint and/or adjoining levels is often required in such cases.[7] Hemilaminectomy and unilateral facetectomy may be options that preserve spinal stability, but yield inadequate exposure of large intradural lesions.

The anterior approach used in isolation to access intradural tumors of the cervical spinal cord has rarely been reported in the literature. O'Toole et al[5] described a case of a ventrally located intradural extramedullary schwannoma at C4-C5 which was resected through an anterior corpectomy approach. This lesion was very similar to the present case. The authors employed post-operative lumbar CSF drainage to prevent an anterior CSF fistula. Epidural bleeding was not found to be significant. Kaya and colleagues described partial removal of a dermoid cyst though a "fenestra corpectomy" in the midpoint of fused cervical vertebrae of a patient with Klippel-Feil.[16] In this case brisk epidural bleeding was encountered. Due to the limited exposure dural closure was not water tight and a pseudo-meningocele was documented on MRI. Owing to the limited corpectomy spinal arthrodesis was not required. In addition, Iwasaki et al described use of the anterior approach for resection of dumbbell shaped tumors in the cervical spine of four patients.[17] In those patients where an intra-dural component required resection, the dura was opened and total resection was achieved. In another article these authors also described an anterior approach to resection of an intra-medullary hemangioblastoma.[18] This tumor was located in the anterior aspect of the spinal cord and thus the anterior approach was favored by these authors. Hakuba et al[19] described an anterior-lateral partial vertebrectomy approach for dumbbell tumors which they called a transuncodiscal approach. This involved a lateral incision of the dura to access the intradural component of these tumors. The exposure involved exposure and retraction of the vertebral artery exposing it to injury which the authors saw as a possible disadvantage. Their case series involved some large tumors which necessitated hypoglossal nerve section and accessory nerve in one case.

Anterior approaches have also very rarely been used to treat anterior intradural pathology other than tumors. Hida et al[20] described five cases of peri-medullary arteriovenous fistulas arising in the region of the anterior spinal artery which were treated using a corpectomy approach. These authors indicated that the intra-operative exposure was limited and that dural closure was not easy. They treated patients with lumbar drainage to avoid post-operative CSF leak successfully in all cases. They favored this surgical technique as it most directly approached lesions on the anterior surface of the spinal cord

The case we have presented did not seem approachable with posterior or posterolateral approaches. The tumor was located ventrally in the midline and the spinal cord was displaced posteriorly but was not displaced laterally or rotated (Figure 2). Therefore, the tumor was anterior to the dentate ligaments as well as all nerve roots. The posterior approach would have required some retraction or rotation of the spinal cord. Furthermore, given the tumor's location bilateral access may have been required to ensure a complete resection. It was largely for these reasons that we chose an anterior approach.

Upon undertaking operation we found that wide vertebrectomy (Figure 3,4) allowed full exposure of the tumor and its attachments. The operative corridor was very adequate and we were able to achieve a full ventral exposure of the spinal canal without any obstructing structures (as opposed to the posterior approach). Dural opening and repair was easily accomplished. This lesion received a vascular pedicle close to the anterior midline which was identified and taken early in the resection. This would not have been accessible until late in the resection through a posterior approach. Moreover, this lesion arose from a ventral rootlet which was also directly approached through this exposure.

Cervical spine stabilization was achieved using fibular allograft to span the defect and anterior cervical plating (Atlantis Anterior cervical plating system, Medtronic) augmented with an external cervical orthoses. Adequate anterior exposure for a lesion such as this tumor requires vertibrectomy and likely multilevel exposure. Long strut graft such as that used in our case, are known to be vulnerable to graft migration, displacement, angulation, fracture, nonunion, and instrumentation failure requiring revision.[21] Anterior plating reduces the risk of graft dislodgement and for exposures with greater than two–level vertibrecomy additional posterior stabilization is recommended.[22,23] At minimum given the high graft related complication rate in these constructs careful follow-up with radiological evaluation should be undertaken.

A concern with the anterior approach has been bleeding from the epidural venous plexus.[5] We found this to be a problem with this approach. We encountered significant bleeding from these veins at the time of initial dural exposure (completion of the corpectomies) and after tumor removal. Clearly at both these intervals pressure upon the veins was released. We were able to achieve hemostasis with gelatin sponge (Gelfoam, Pfizer) and microfibrillar collagen (Instat, Ethicon). Due to the brisk blood flow we found gelatin sponge to be superior in this situation. Strategies to control intraoperative bleeding can include techniques such as hypotensive anesthesia, patient positioning (head up, avoidance of increased venous pressure through abdominal compression or increased thoracic pressures), use of systemic antifibrinolytics, local trombotic agents and blood salvage techniques (e.g. cell saver). These strategies have been recently reviewed in the context of complex spinal surgery.[24,25] Preoperative angiography is commonly used in the setting of hypervascular metastatic tumors but has not been commonly applied to intradural tumors.[26] In our case the majority of the blood loss appeared to come from compressed and likely dilated epidural veins. These present a particular challenge as one must be careful to avoid compression of the thecal sac when applying cautery or local hemostatic agents. One possible strategy that may have assisted in our case would be to attempt removal of the tumor "piece-meal" in order to avoid sudden decompression of the epidural venous plexus.

Another concern has been postoperative CSF fistula due to the difficulty achieving watertight suture of the dural incision and due to a large postoperative dead space. We encountered no difficulties in suturing the dura in a running fashion using braided nylon suture (Nurolon, Ethicon).

In conclusion intradural extramedullary tumors in the cervical spine are usually approached through a posterior or posterolateral approach. Resection through an anterior corpectomy is an alternative not often employed. This approach is suited for resection of tumors which are located in the ventral midline. It provides excellent visualization of these lesions. Epidural venous bleeding can be problematic. In situations of rapid blood loss gelatin sponge may be more effective than microfibrillar collagen because of its ease in handling and tamponading abilities. Dural closure and a limited operative corridor should be considered by the surgeon but were not found restrictive in our hands.

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