Original Article

Surgical approach to C1-C2 nerve sheath tumors

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Background: C1 and C2 nerve sheath tumors (NST) are unique in presentation, relationship to neighbouring structures and surgical approaches when compared to their counterparts in other regions of the spine. **Aim:** The strategies involved in the surgery for C1-C2 NST are discussed **Setting and Design**: Retrospective study. **Methods:** 21 patients with C1 (n=6) and C2 (n=15) NST were operated based on their position with respect to the cord i.e. anterior (4), anterolateral (10), posterolateral (5), and posterior (2). The tumors had extra- and intradural components in 20 patients; while in one, the tumor was purely intradural. The operative approaches included the extreme lateral transcondylar approach (3); laminectomy with partial facetectomy (5); laminectomy (11); and, suboccipital craniectomy and laminectomy (2). **Results:** Total excision was performed in 13 patients; while in 7, a partial extraspinal component, and in 1, a small intradural component were left, in situ. Thirteen patients showed improvement by one or more grades in the Harsh myelopathy score; 2 patients with normal power had significant decrease in spasticity; while 5 maintained their grade. One poor-grade patient succumbed to septicemia. **Conclusions:** C1-C2 NST may have exuberant growth due to the capacious spinal canal and the absence of a "true" intervertebral foramen at this level. Surgical approaches are determined by its relationship to the cord . A "T incision" on the dura, the partial drilling of the facets, sectioning of the denticulate ligament, rotating the operating table 15 to 30 degrees, and at times sectioning the posterior nerve roots are all useful adjuncts for facilitating access.

Key Words: Spinal nerve sheath tumors, craniovertebral junction, laminectomy, extreme lateral transcondylar approach

Introduction

Nerve sheath tumor (NST) of the C1 and C2 roots have certain characteristic features not shared by its counterparts in the other regions of the vertebral column.[1,2] The surgical management of this lesion depends upon its location with respect to the spinal cord and nerve roots; the extent of its extra-dural and extra-spinal extension; the relationship of the tumor to the vertebral artery; and, the presence of multiple NST in the cervical spine.[3-8] The unique osseous anatomy and the biomechanical features of this region also merit special considerations.[9] In the present study, the strategies involved in the surgery for C1-C2 NST are being discussed.

Materials and Methods

Clinical spectrum

In this retrospective study, 21 patients (16 men and 5 women, aged 17-56 years, mean age 34.8 +12.7 years) operated for C1-C2 NST between 1st January 1994 and 31st December 2002, were studied. The C1-2 NST accounted for 12.1% (21out of 173) and 33% (21 out of 63) of all spinal and cervical NST, respectively. Their duration of symptoms ranged from 3 to 60 months (mean duration=21.8 $\pm$19.5 months). The symptoms at the onset of illness were spasticity (n=10, 47.6%), tingling and numbness below neck (n=4,19.0%), weakness (n=3,14.3%) and neck pain (n=4,19.0%). Their clinical presentation is summarized in Table 1. Two patients in the series had neurofibromatosis. One of these patients, with a C1-C2 NST and bilateral acoustic schwannomas, had papilledema and IX-Xth cranial nerve palsy in addition to facial hypoaesthesia. An associated neurofibroma at the L_4 level was present in the other patient. One patient presented with a swelling in the neck behind the posterior border of the sternocleidomastoid due to extension of the extraforaminal component of the lesion.

Grading

The clinical disability of the patient was evaluated using the Harsh Myelopathy Grading [10] (Table 2) as: Grade 1: normal power with only hyperreflexia and Babinski's sign (n=2, 9.5%); Grade 2: able to walk but not able to run (n=5, 23.8%); Grade 3: requiring support to ambulate (n=11, 52.4%) and Grade 4: completely bedridden (n=3,14.2%).

Radiological findings

The patients were evaluated using lateral radiographs of the

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Table 1: Clinical presentations of the 21 patients with C1-C2 nerve sheath tumors

Presenting Features	Number of patients	Percentage
Spasticity	21	100
Weakness	18	85.7
Spinothalamic tract involvement	11	52.4
Posterior column involvement	15	71.4
Horner's syndrome	5	23.8
Sphincter dysfunction	6	28.6
Dyspnea at rest	3	14.3
Neck pain	10	47.6
Neck tilt	5	23.8
Restriction of neck movements	7	33.3
Suboccipital muscle wasting	4	19
Facial hypoaesthesia	4	19
Nystagmus	4	19
Papilledema	1	4.8
IXth-Xth nerve palsy	1	4.8
Neck swelling	1	4.8

Table 2: Pre-and postoperative Harsh Myelopathy Grade [13] of the patients

Harsh Grade	Preoperative	Postoperative
1	2	3
2	5	16
3	11	1
4	3*	-

*one patients expired after surgery.

craniovertebral junction (CVJ) and magnetic resonance imaging (MRI). In our series, only 1 patient had a purely intradural tumor while the rest had both intra-and extradural components. For a preoperative planning, the relationship of the tumor with the cord was determined with the help of MRI and classified into 4 types—anterior, anterolateral, posterolateral and posterior (Table 3, Figure 1). Four (19%) of the tumors in the present series were anterior (Figure 2 a, b, and c); 10 (47.6%) anterolateral (Figure 3 a, b, c; 4 a and 4 b); 5 (23.8%) posterolateral; and, 2 (9.5%) posterior. On T1 weighted MR sequences, the lesions were hypointense in 10 and isointense in 11 patients. They were heterogeneously hyperintense in all patients on T2 sequences. Eleven (52.4%) had hyperintense signal changes of the cord at the level of the tumor. The plain radiographs of the cervical spine revealed scalloping of vertebral bodies in 5, widening of interlaminar space in 4, and both in 4 cases. The plain radiographs were normal in 8 patients.

Surgical procedures (Table 3)

Of the 4 patients with anteriorly located tumors, 3 underwent total excision using the extreme lateral transcondylar approach.[11,12,13] In 1 patient with a similar anteriorly placed tumor in whom the tumor was extending above the foramen magnum (FM) and laterally along the nerve root, the tumor was accessible using a suboccipital craniectomy and C1-C2 laminectomy.

The commonest location was the anterolateral one (n=10). In 5 of these patients, a laminectomy with partial facetectomy[3] provided an oblique access to the tumor, while in 5 others, only a laminectomy sufficed. In all the 10 patients, a T-incision was given on the dura in order to connect the intra- and extradural components. Access to the part of the lesion located anterior to the cord within the intradural space was greatly facilitated by excision of the denticulate ligament.

The posterior and posterolaterally located lesions were approached by a laminectomy alone. In one patient, however, in whom the lesion was extending above the foramen magnum, a suboccipital craniectomy was also performed.

The NST were arising from C1 and C2 roots in 6 (28.6%) and 15 (71.4%) patients, respectively. One or more rootlets were sacrificed in all cases. In 9 cases, the entire nerve root from which the tumor was arising was sacrificed. A patch graft was used to close the dura in 11 cases (in 1 patient who underwent an extreme lateral transcondylar approach; in 5 patients who underwent laminectomy and partial facetectomy; in 4 patients who had a laminectomy with a 'T' incision on the dura; and, in 1 patient who underwent a suboccipital craniectomy and laminectomy). It was possible to primarily close the dura without a patch in rest of the cases.

Table 3: Location of the lesions relative to the spinal cord and the surgical approach adopted in them

Location	Number	Surgical Approach			
		ExLTcA*	SOC+L•	L+F**	L#
Anterior	4	3	1	-	-
Anterolateral	10	-	-	5	5
Posterolateral	5	-	1	-	4
Posterior	2	-	-	-	2

Abbreviations used: *Extreme lateral transcondylar approach; • suboccipital craniectomy and laminectomy; ** laminectomy with partial facetectomy; # only

Results

Total excision was achieved in all patients except in 6 of the 10 (60%) anterolaterally placed and 2 of the 5 (40%) posterolaterally placed lesions. In 7 of these patients, a part of the tumor extending far laterally beyond the confines of the spinal canal was not removed; and, in 1 patient with an anterolaterally placed lesion, a small part of the lesion located anterior to the cord that did not have a good plane of cleavage, was left, in situ.

At clinical assessment performed at the time of discharge, 13 of the 21 patients showed improvement by one or more grades while 7 maintained their status. The pre- and postoperative disability grades are summarized in Table 2.

sOne of our patients with an anteriorly situated NST developed sudden quadriplegia and respiratory arrest prior to sur-

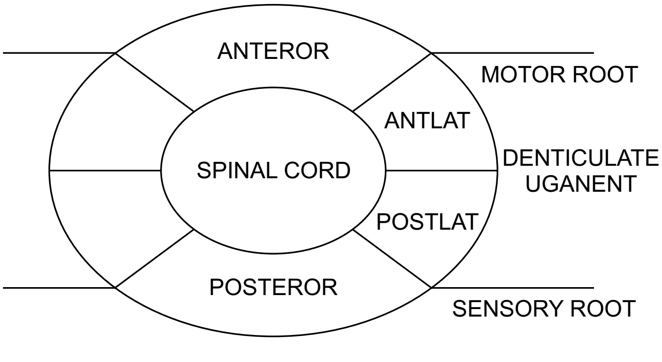

Figure 1: Classification of tumors with respect to their location relative to the cord, roots and the denticulate ligament.

320 CMYK

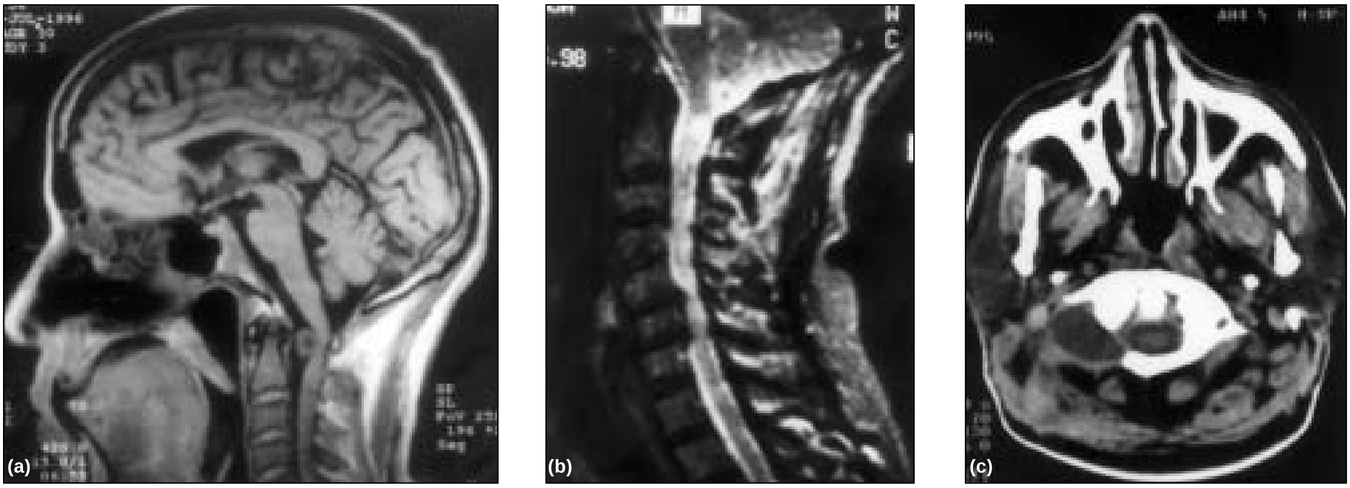

Figure 2: (a) T1-weighted sagittal MR image showing a nerve sheath tumor located anterior to the cord opposite the odontoid causing cervicomedullary compression; (b) T2- weighted MR image showing complete tumor removal using an extreme lateral transcondylar approach. Signal intensity changes are still persisting in the cord opposite the original site of lesion; and, (c) Axial CT image showing the resection of part of the C1 facet and total tumor removal.

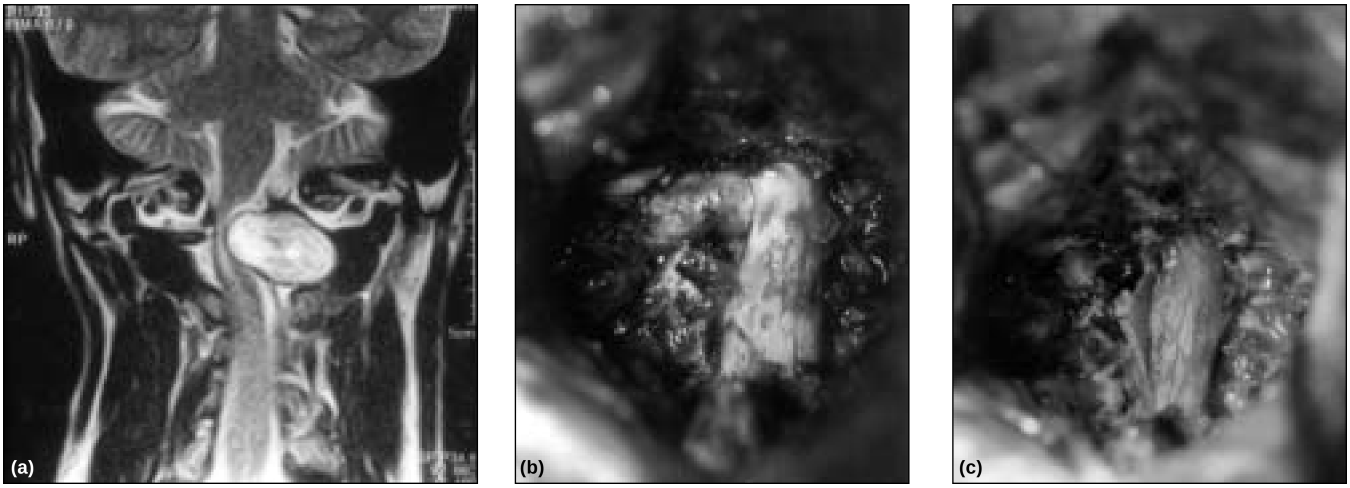

Figure 3: (a) Preoperative T2-weighted coronal MR image showing a dumb-bell nerve sheath tumor with a large extraspinal component; (b) Intraoperative photograph of the C2 neurinoma exiting through the intervertebral 'foramina'; and, (c) Intraoperative photograph showing total excision of the intra- and extradural component of the tumor.

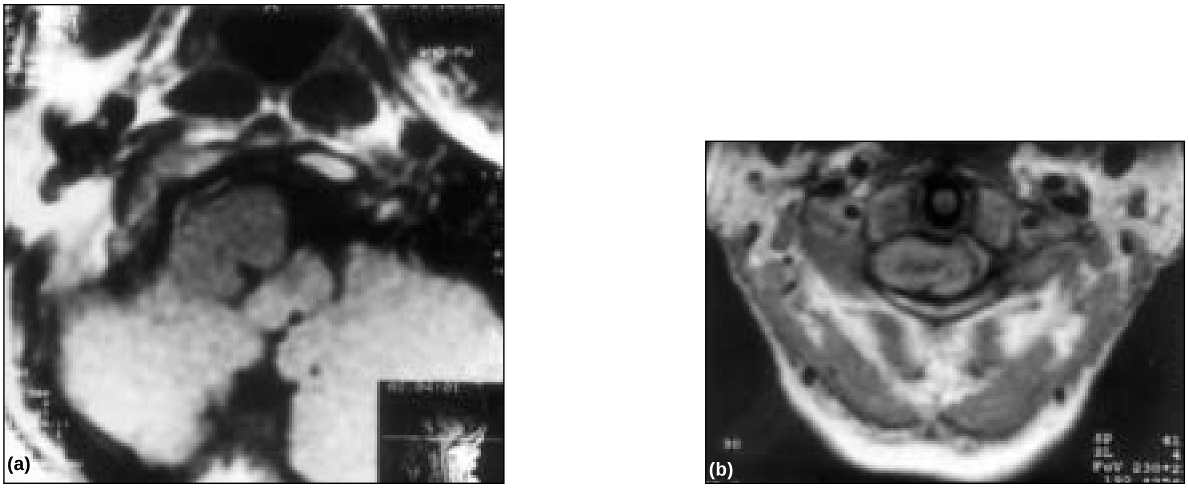

Figure 4: (a) T1-weighted axial MR image showing an anterolaterally placed tumor extending above foramen magnum pushing the cord posterolaterally; (b) T1-weighted axial MR image showing minimal extraspinal extension.

gery that did not improve despite total excision of the tumor. He succumbed to bronchopneumonia and septicemia. The two patients (where a patch dural graft was placed) who developed CSF leak from the operative incision were managed by acetazolamide administration and lumbar drainage.

Discussion

Incidence

C1-2 NST represent approximately 5% of spinal and 18% of cervical NST reported[2,7,14,15] while in the present series, they accounted for 12.1% and 33% of the spinal and cervical NST, respectively. Of these, 15 (71.4%) originated from the C2 root and 6 (28.6%) from the C1 root, respectively. This is in agreement with the studies published by Yasuoka, et al,[7] with 18 tumors on C2 and 1 on C1 root; by Guidetti et al,[15] with 6 on C2 and 3 on C1; and, George et al,[2] with 44 on C2 and 6 on C1, respectively. The latter have also reported multiple lesions due to a high incidence of neurofibromatosis in their series.[2] In our study, the two patients with neurofibromatosis had only solitary lesions of the cervical spine. One of these patients had associated bilateral acoustic schwannomas, while the other had a NST within the lumbar spine.

Anatomical considerations

In the present series, 20 patients had dumb-bell lesions with both extra- and intradural extensions, while only one had a purely intradural location. Similarly, George et al[2] have found 83% of C1-2 nerve sheath lesions having an extradural extension. In contrast, the total incidence of extradural extension of NST at other levels within the spine is only 16%.[2]

The relatively large size of these lesions at the C1-2 level is due to the spacious spinal canal at this level. The mean sagittal diameters at the levels of the atlas and axis are 23 and 20mm, respectively,[9] while in the subaxial spine, the average diameter is only 15mm.[9] The cervical bulge of the spinal cord also begins below the axis.[9] At the occipitocervical and atlanto-axial levels, the facetal pillars lie anterior to the nerve roots exiting through the intervertebral foramina. This also permits an exuberant growth of the lesion outwards since there is no posterior bony obstruction to its growth at the C1-2 levels.[9] There is often a delay in the development of clinical symptoms until these tumors attain a fairly large size.[4,9]

Surgical approaches (Table 4)

The MR findings (Figure 1) were utilized to determine the surgical approach. The posteriorly located lesions were accessed using a laminectomy. In the cases of multi-segment involvement, laminoplasty and hemilaminectomy[16] avoid the post-laminectomy kyphosis.[7,12] The posterior approach also accessed the anterolateral or posterolateral lesions that, having shifted the cord towards the contralateral side, extended laterally towards the intervertebral foramen along the nerve roots. The intra-tumoral decompression of the lateral part of the tumor provided an oblique trajectory to access the part of the lesion situated in close proximity to the cord. In our series, sectioning of the denticulate ligament,[17] rotating the operating table 15 to 30^0 and at times, sectioning the posterior nerve roots, as suggested by Bucci et al,[1] in all anterolateral lesions and the one anteriorly placed lesion treated by the posterior approach, aided in gently mobilizing the tumor from the contact-surface of the cord. Placement of a “T” shaped dural incision, with the horizontal limb of the “T” connecting the intra- and extra-dural components of the tumor also facilitated access.

In the case of dumb-bell tumors, initially an intratumoral decompression was performed. Then, the intradural portion of the lesion was removed maintaining arachnoidal planes in the tumor-cord interface to avoid traction on the cord during the handling of the extradural portion. The use of operative microscope and neurophysiological monitoring facilitate tumor excision with minimal cord manipulation.

In the present series, the anatomical constraints posed by the posterior approach in accessing anterolaterally located lesions were overcome by extending the surgical corridor laterally by adopting a posterolateral trajectory via a laminectomy and partial facetectomy, as proposed by Bartolomei and Crockard.[3] This approach is also advantageous in gaining an early control of the vertebral artery.[3]

Does laminectomy with partial facetectomy lead to cervical instability?[16, 18,19] In the two series by George and Lot,[2,14] of cervical NST, as well as the study by Bartolomei and Crockard[3] of bilateral C2 neurofibromas, none of the patients have required fusion for cervical instability after laminectomy and partial facetectomy. At the level of C1-2, the anterior ligaments confer an additional stability to the cervical spine with less structural contribution from the posterior spinal column.[3] The partial preservation of bilateral articular facets between C1 and C2 also confers stability. According to Welling et al,[9] C1-C2 lateral joints have a capsule that is lax on the medial side. Most of the movement occurs along a synovial fold present medially. Removal of this part, therefore, does not lead to instability. In our patients with anteromedially placed NST, only a unilateral partial facetectomy with preservation of more than two –thirds of the joint surfaces and capsule was performed and the contralateral joint was left completely intact. Thus, no spinal fusion was required.

For anterolaterally placed lesions, Verbeist[20] and George et al,[2] use an approach that partially removes the anterior portion of the vertebral body and transverse foramen but preserves the facet. The vertebral artery is mobilized from the foramen transversarium and the extraspinal tumor can also be safely excised. However, the extensive drilling and the risk of vertebral artery mobilization for resecting essentially the extraspinal component of the benign lesion (that was unlikely to cause spinal cord compression even in the distant future)

Table 4: Review of the approaches to C1-C2 nerve sheath tumors

Approaches	Advantages	Disadvantages
Anterior [22,23] [transoral approach]	Direct access to tumor with no cord retraction	Greater risk of infection Watertight dural closure is difficult Lateral extensions of the tumor cannot be accessed.
Anterolateral[14,20]	Total removal of extraforaminal component possible Arterial supply to the tumor can be coagulated before its decompression in order to reduce its vascularity.	Need for vertebral artery mobilization Troublesome bleeding from the venous plexus can occur. Not ideal for cases above the axis vertebra due to the obliquity of the approach Removal of the intradural component requires an additional laminectomy or hemicorpectomy. Postoperative Horner's syndrome may be a sequel
Posterolateral [3] [Laminectomy and medial ipsilateral partial facetectomy]	Removal of the medial half of facetal pillars gives excellent exposure. No roots or vessels intervene in the path. Root sacrifice, denticulate ligament sectioning, "T"incision on the dura all contribute to increasing the field of view in this approach.	Risk of instability. Extraspinal component not adequately accessed.Hence recurrence may occur. Pole of tumor in intimate contact with vertebral artery may not be visible to the surgeon.
Lateral [extreme lateral transcondylar approach] [11,12,13]	Direct visualization of anterior craniovertebral junction Removal of both extra - and intradural components is facilitated. Access to extension of the tumor above the foramen magnum is possible Interface between the tumor and the anterior surface of cord is well made out Lateral opening of the dura avoids the force-vector that may cause excessive mobilization of the cord in true anterior/anterolateral lesions.	Unfamiliar anatomy for most surgeons Bilateral tumors cannot be dealt with in the same sitting Need for stabilization if more than one third of the occipital condyle is drilled. Post-operative scarring makes it unsuitable for tackling recurrences
Posterior [6,8] [standard bilateral laminectomy with or without suboccipital craniectomy]	Simplest procedure Lesser risk of instability.	Inadequate access to anteriorly/ anterolaterally placed tumors No access to extraforaminal tumor

prevented us from adopting this approach.

In patients with lesions situated mainly anterior to the cord, the extreme lateral transcondylar approach was used.[11,12,13] Though an early control of the vertebral artery, a direct visualization of the cervicomedullary junction and the cord-tumor interface from a lateral aspect, and a field of view above and below the foramen magnum were obtained, the extensive bony and soft tissue dissection were the major deterrents to its frequent use in all dumb-bell, anteriorly placed lesions. The extrapharyngeal approach to the upper cervical spine may be an alternative approach.[21] The transoral approach for midline, anterior, intradural NST[22,23] has the risk of infection due to traversing through a potentially infected oral cavity;[24] and, is unable to access tumors extending laterally from the midline.

Implications of the C1/ C2 nerve root resection

In removing the spinal NST, preservation of the anatomical integrity of the involved nerve root, although desirable, cannot always be achieved. This is specially in cases of large tumors; where both the sensory and motor roots are diffusely involved by the pathological tissue; and, when the tumor extends beyond the proximal pole of the dorsal root ganglion, where the roots are devoid of an arachnoidal sheath that usually separates them from the tumor tissue.[4, 25,26] Occasionally, the posterior sensory nerve root may be sacrificed in order to gain access to the anteriorly located tumor.

In our series, one or more rootlets were sacrificed in every case, while in 9 cases, the entire nerve root, from which the tumor arose, was resected. As the C1 and C2 nerve roots are nonappendicular and therefore relatively non-eloquent, this was not associated with any significant neurological deficit except an occasional mild hypoaesthesia in the suboccipital region.[26, 27,28] While working with eloquent nerves in the subaxial spine, Lot and George[14] minimized postoperative deficits by electrically stimulating the nerve during surgery. When a response was recorded, the functional root was preserved even at the cost of leaving some residual lesion. This procedure was not required in our study since the resection of C1 or C2 root is usually not associated with any significant sensory or motor radicular symptoms.

Implications of incomplete removal of the extra-spinal tumor

In 7 patients, a part of the extraspinal tumor extending far laterally beyond the intervertebral foramen was not removed. Usually this part of the tumor has neither clinical recurrence nor a tumor regrowth into the spinal canal even after extremely long intervals ranging from 6 to 18 years. Preserving the nerve roots at the cost of a subtotal tumor removal is also not associated with increased recurrence rates.[5] Seppala, et al,[29,30] have reported that of the 187 patients treated for spinal NST, in the 20 in whom less than total tumor excision was performed to avoid root injury, 18 did not have a symptomatic recurrence even after a median period of 19 years after surgery. In the patients with neurofibromatosis, the recurrence rates in the cases of partial tumor excision are higher.[28,29,30] However, only two of our patients had neurofibromatosis and in both of them, a total tumor excision could be achieved.

In conclusion, C1 and C2 nerve sheath tumors are special when compared to their counterparts in other regions of the spine in terms of their tendency toward extradural and extraspinal spread, relationships to neighbouring structures and the surgical approaches required to deal with them. They were classified based on their position with respect to the cord determined by preoperative magnetic resonance imaging. A "T incision" on the dura, the partial drilling of the facets, sectioning of the denticulate ligament, rotating the operating table 15 to 30 degrees, and at times sectioning the posterior nerve roots are all useful adjuncts for facilitating access to these lesions.

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