Original Article

Surgical management of spinal schwannomas arising from the first and second cervical roots: Results of a cumulative case series

ABSTRACT

Objective: Schwannomas of the first and second nerve roots are rare neurosurgical entities, harboring specific surgical features that make surgical resection particularly challenging and deserve specifics dissertations. This study is a retrospectively analysis of 14 patients operated in two different neurosurgical centers: the San Filippo Neri Hospital of Rome and the Federal Centre of Neurosurgery of Tjumen.

Materials and Methods: In the last 6 years, 14 patients underwent neurosurgical resection of high cervical (C1–C2) schwannomas, in two different neurosurgical centers. Patients data regarding clinical presentation, radiological findings, and surgical results were retrospectively analyzed.

Results: The mean age was 50 years (range 13–74), the follow-up mean duration was 30 ± 8.5 (range 24–72 months), and there was no significant differences among different tumor locations (intradural, extradural, and dumbbell). Surgical results were excellent: gross total resection was achieved in all cases and there were no intraoperative complications or postoperative mortality. All patients presented postoperative clinical improvement except one who remained stable. Karnofsky performance status, at the last follow-up, confirmed a global clinical improvement. No vertebral artery (VA) injury neither spinal instability occurred; nerve root sacrifice was reported in one case.

Conclusions: Neurosurgical treatment of C1–C2 schwannomas is associated with good outcomes in terms of extent of resection and neurological function. In particular, dumbbell shape and VA involvement do not represent limitations to achieve complete tumor resection and good clinical outcome. In conclusion, microsurgery represents the treatment of choice for C1–C2 schwannomas.

Keywords: C1–C2 schwannomas, craniocervical junction, dumbbell tumors, spinal schwannomas, vertebral artery

INTRODUCTION

Spinal schwannomas constitute more than one-third of all primary spinal tumors.[1-5] However, schwannomas arising from the first two cervical nerve roots are very rare and only few reports described series of C1 and C2 schwannomas separately.[6-10] Indeed, high-cervical schwannomas harbor specific features that make surgical resection particularly challenging. Nonetheless, gross total resection (GTR), preserving neurological functions, appears to be the preferred treatment to minimize recurrence risks.[2,11] We report the results of a cumulative series of C1–C2 schwannomas and discuss clinical data, radiological findings, and surgical results of 13 patients treated in two different centers of neurosurgery.

Francesco Corrivetti[1,2], Raffaelino Roperto[1], Rinat Sufianov[3,4], Guglielmo Cacciotti[1], Arslan Musin[3], Albert Sufianov[3,4], Luciano Mastronardi[1,4]

[1]Department of Neurosurgery, San Filippo Neri Hospital, ASL Roma 1, Rome, [2]Laboratory of Neuroanatomy, EBRIS Foundation, Salerno, Italy, [3]Federal Centre of Neurosurgery, Tyumen, [4]Department of Neurosurgery, The State Education Institution of Higher Professional Training, The First Sechenov Moscow State Medical University Under Ministry of Health, Moscow, Russia

Address for correspondence: Dr. Luciano Mastronardi, Via Reno 14, Roma 00198, Italy.
E-mail: mastro@tin.it

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MATERIALS AND METHODS

Data collection

We retrospectively reviewed a consecutive series of 31 craniocervical junction (CCJ) schwannomas, operated, in the last 6 years, in two different neurosurgical centers: the San Filippo Neri Hospital of Rome and the Federal Centre of Neurosurgery of Tjumen. Clinical data were collected and shared in a common institutional anonymized electronic medical database. The records were retrospectively analyzed regarding clinical presentation, radiographic assessment, surgical outcome, and follow-up period [Table 1]. This retrospective case series was reviewed and approved by the institutional review board of both hospitals. Written informed consent was obtained for surgery. Considering that the study was retrospective, written consent to participate in the study was not applicable.

Tumor classification

Tumors were classified in intradural, extradural, and dumbbell shape [Figure 1a-c]. The vertebral artery (VA) tumor involvement was evaluated as spared (seven cases), compressed (four cases), or encased (two cases) by tumor involvement [Figure 1 d-f]. Dumbbell tumors were also classified, according to the Eden classification[12] [Figure 2], based on the presence or absence of the intradural portion (Eden Type A or B and Eden Type C or D). Tumor location within the spinal canal was defined according to the classification of foramen magnum tumors, described by George *et al*.,[13] that categorize extramedullary tumors located between the midline and the dentate ligament as lateral. Nonetheless, schwannomas in this series were further classified in anterolateral or posterolateral, if a clear anterior or posterior extension in the spinal canal was observed. Radiological diagnosis was made for all patients with craniocervical magnetic resonance images (MRI). Computed tomography angiography was performed in selected cases to determine the relationship between tumor mass and VA or to evaluate erosive or compressive bony changes.

Literature review

We performed a literature review of the surgical series of C1–C2 peripheral nerve sheath tumors (PNSTs). In order to compare previous surgical results to the present study, case series of C1–C2 PNSTs presenting exclusively dumbbell shaped tumors were excluded. We found eight papers corresponding to the required criteria. Only four of them reported specific C1–C2 case series.[7-10] Two of them focused on CCJ tumors,[14,15] and the other two reported series of tumors located at high cervical level (C1–C3).[6,16] Each study was analyzed to extrapolate differences in tumor location, extent of resection (EOR), complications, recurrence, and mortality rates [Table 2].

RESULTS

Patients data and tumor location

Among the 31 cases of CCJ schwannomas, we excluded all patients with a known diagnosis of neurofibromatosis or those with tumors arising from lower cranial nerves. A total of 13 tumors arising from the first and second nerve roots were included in the study. Among them, seven cases were operated at blinded for review and six cases at blinded for review. The mean age was 50 years (range 13–74); male: female ratio was 8:5. Follow-up period ranged between 24 and 72 months (mean 30 ± 8.5). Pediatric

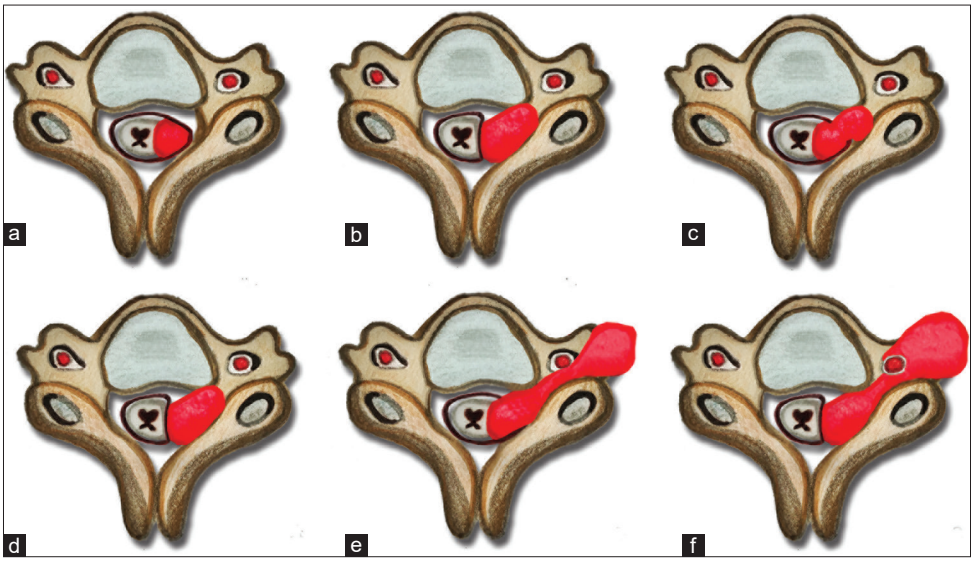

Figure 1: The drawing tumor location and vertebral artery (VA) involvement: (a) Intradural, (b) Extradural, (c) Intra–extradural, (d) VA spared, (e) VA compressed, (f) VA encased

population represented the 7% of entire series (one case). The mean maximal tumor size was 3.7 cm and varied from 2 to 5.1 cm. Schwannomas arose from the C1 nerve root in 3 patients (23%) and from C2 nerve root in 10 patients (77%). Isolated intradural location was reported in two cases, two lesions were exclusively extradural, and nine had a dumbbell shape. Among them, four were Eden Type A, four Eden Type C, and one was Eden Type D. There were in our series two anterolateral, five lateral, and six posterolateral lesions.

Table 1: Summary of the study population

Study population features	
Age (years), mean±SD	50±21
Sex (male %)	8 (61.5)
Postoperative - follow-up (months), mean±SD	30±8.5
Clinical presentation, number of cases (%)	
Cervical pain, headache	8 (57.1)
Sensory deficit upper extremity	5 (35.7)
Sensory deficit lower extremity	3 (21.4)
Motor deficits upper extremity	5 (35.7)
Motor deficits lower extremity	5 (35.7)
Gait disturbance	1 (7.1)
Tumor size (mean cm)	3.7
Nerve root involvement, number of cases (%)	
C1	3 (23)
C2	10 (77)
Tumor location, number of cases (%)	
Intradural	2 (15.3)
Extradural	2 (15.3)
Dumbbell	9 (69.4)
VA involvement, number of cases (%)	
Spared	7 (53.8)
Compressed	4 (30.7)
Encased	2 (15.3)

SD - Standard deviation; VA - Vertebral artery

Clinical presentation

The main clinical presentation at diagnosis consisted of headache, neck, or shoulder pain in 57% of the cases ($n = 8$). Numbness and paresthesia occurred in five cases for upper extremity and in three cases for lower extremity. Motor deficits were equally present in the upper and lower extremities in five cases. Gait disturbance occurred in one patient. The Karnofsky performance status (KPS) was administered to each patient at admission, discharge, and follow-up [Table 3].

Surgical approaches

Several surgical approaches, performed by the senior authors (LM and AS), have been utilized in this case series. The majority of patients ($n = 11$) were treated through a posterior cervical midline approach and C1–C2 hemilaminectomy ($n = 6$) or C1–C2 laminectomy ($n = 5$) [Table 4]. Far-lateral approach was preferred in two cases of respectively extradural and dumbbell tumor located at C1 with lateral extension. All

Table 2: Literature review

	Nerve root involvement				Tumor location			EOR		Outcome		
	C1	C2	Multiple	Total	Intradural, *n* (%)	Extradural, *n* (%)	Intra–extradural, *n* (%)	GTR, *n* (%)	STR-PR, *n* (%)	Complications, *n* (%)	Recurrence, *n* (%)	Mortality, *n* (%)
Guidetti *et al*. (1988)	3	6	0	9	NR	NR	NR	6 (100)	0	2 (22.2)	0	1 (11.1)
George and Lot (1995)	5	30	7	42	7 (17)	16 (38)	19 (45)	37 (88)	5 (12)	2 (4.7)	2 (4.7)	2 (4.7)
Krishnan *et al*. (2004)	6	15	0	21	1 (5)	0	20 (95)	13 (62)	8 (38)	2 (9.5)	NR	1 (4.7)
Maurya *et al*. (2009)	10	18	4	32	2 (6)	10 (31)	20 (63)	26 (81)	6 (19)	2 (6.2)	2 (6.2)	1 (3.1)
Watanabe *et al*. (2009)	NR	NR	0	9	1 (11)	0	8 (89)	NR	NR	2 (22.2)	1 (11.1)	0
Wang *et al*. (2011)	2	16	0	18	0	4 (22)	14 (78)	15 (83)	3 (17)	6 (33.3)	1 (5.5)	1 (5.5)
Cavalcanti *et al*. (2011)	NR	NR	0	13	5 (38)	4 (31)	4 (31)	13 (100)	0	2 (15.3)	0	0
Chowdhury *et al*. (2013)	2	11	0	13	8 (62)	2 (15)	3 (23)	15 (100)	0	2 (15.3)	0	1 (7.6)
Present study	4	10	0	14	2 (15)	2 (15)	9 (70)	13 (100)	0	0	0	0

EOR - Extent of resection; GTR - Gross total resection; STR-PR - Subtotal-partial resection; NR - Not reported

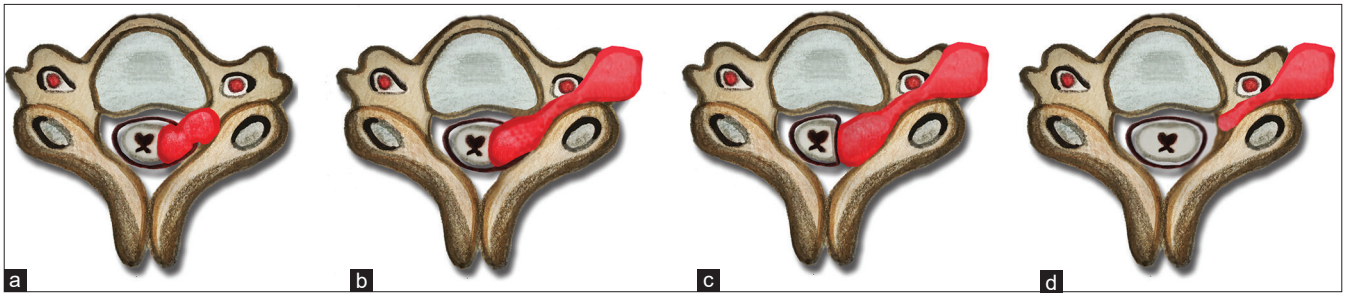

Figure 2: Illustration of Eden's classification of dumbbell tumors. (a) Type I, intradural and extradural, (b) Type II, intradural, extradural, and paravertebral, (c) Type III, extradural and paravertebral, (d) Type IV, foraminal and paravertebral

Table 3: Summary of radiological findings, surgical approach, and clinical outcome of the study population

Case number	Age, sex	Nerve root involvement	Intraspinal location	Intradural extradural dumbbell	Eden class*	VA involvement	Surgical approach	KPS preoperative	KPS postoperative	Outcome	Complications	F/U
1	13, male	C1	Lateral	Extradural	-	Spared	Far-lateral approach	60	80	Improved	None	24
2	45, female	C2	Posterolateral	Dumbbell	D	Compressed	C2 hemilaminectomy	90	100	Recovered	None	42
3	49, male	C2	Posterolateral	Intradural	-	Spared	C2 laminectomy	60	90	Improved	None	36
4	56, female	C2	Anterolateral	Dumbbell	A	Compressed	C2 laminectomy	60	90	Improved	None	48
5	70, male	C2	Lateral	Dumbbell	C	Compressed	C2 hemilaminectomy	60	90	Improved	C1 nerve root sacrifice	60
6	74, female	C2	Lateral	Dumbbell	C	Encased	C2 laminectomy	90	100	Recovered	none	36
7	65, female	C1	Lateral	Dumbbell	A	Spared	Far-lateral approach	90	100	Recovered	None	74
8	74, male	C1	Lateral	Dumbbell	A	Spared	C1 hemilaminectomy	90	100	Recovered	None	36
9	38, female	C2	Posterolateral	Extradural	-	Spared	C2 hemilaminectomy	90	100	Recovered	None	60
10	20, male	C2	Posterolateral	Dumbbell	C	Encased	C2 hemilaminectomy	90	100	Recovered	None	48
11	60, female	C2	Posterolateral	Dumbbell	A	Spared	C1 hemilaminectomy and C2 laminectomy	90	100	Recovered	None	36
12	53, male	C2	Posterolateral	Dumbbell	C	Compressed	C1 hemilaminectomy and C2 laminectomy	80	100	Recovered	None	42
13	73, male	C2	Anterolateral	Intradural	-	Spared	C2 hemilaminectomy	60	90	Improved	None	24

*Eden classification for dumbbell tumors. F/U - Follow-up (months); KPS - Karnofsky performance status; VA - Vertebral artery

procedures were executed without the necessity of fixation for instability.

Intraoperative VA involvement was managed as follows:

a. Tumor compressing the VA ($n = 4$) was operated without proximal or distal VA control, debulking the tumor, after exposure of posterior surface, following the plane between the VA and the tumor capsule
b. Tumor encasing VA ($n = 2$) was operated using proximal arterial control and subsequent extraperiosteal dissection of the VA as described by George *et al*.[17]

Surgical results

GTR was accomplished in all cases and confirmed at postoperative contrast-enhanced MRI performed between 1 and 3 months after surgery. There were no intraoperative complications, postoperative mortality, or severe deficits. Minor complications as cerebrospinal fluid leakage or spinal instability did not occur. Neurophysiological monitoring, including somatosensory evoked potentials, was used in most cases. Nerve root was sacrificed only in one patient with C1 tumor without clinical consequences. There were not VA injuries in the series. Histological examinations revealed that all tumors were benign schwannomas (WHO grade I), and therefore none of the patients underwent postoperative radiotherapy. At follow-up, ranged between 24 and 72 months (mean 30 ± 8.5), no recurrences were observed, and a global clinical improvement was obtained in all cases, among them eight patients experienced complete recovery.

DISCUSSION

Schwannomas are the most frequently encountered PNSTs, accounting for more than one-third of all primary spinal tumors.[1-5] They usually arise as a solitary tumor, even if they can be associated with neurofibromatosis Type I or II and presenting as multiple tumors.[18,19] Spinal schwannomas of the first two cervical nerve roots are rare and represent around 5% of all spinal neurinomas.[7,10,20] They are often included in large case series of spinal[4,5,19,21,22] or CCJ[14,15,20] tumors, and only a few reports specifically investigated C1–C2 schwannomas alone.[6-8,16] Nonetheless, schwannomas of the first two nerve roots have specific surgical features as involvement of VA, compression of upper spinal cord or dumbbell shape, that deserve specifics dissertations.

Anatomical considerations

High cervical schwannomas more frequently arise from the second nerve root. George and Lot[7] reported a series of

42 patients, harboring 50 PNSTs (35 single and 7 multiple), in which six originated from C1 and 44 from C2. Wang *et al.*[10] described surgical results of a case series of 18 patients, harboring C1 and C2 schwannomas in 2 and 16 cases, respectively. Similarly, our series is composed of four C1 and ten C2 schwannomas.

C1–C2 schwannomas are frequently associated with large size[23-25] or dumbbell shape compared to the other spinal levels.[11,26-28] This feature is probably related to the anatomical configuration of the C1–C2 spinal segment that presents a large intervertebral space and lacks for the intervertebral foramen allowing schwannomas to exit the spinal canal and growing in the paravertebral space. In fact, while common percentage of dumbbell tumors, in spinal schwannomas series, ranged from 6% to 19%, C1–C2 schwannomas reported a percentage of dumbbell tumors significantly higher, ranging from 43% to 95%.[7,8,10] Indeed, in our series, the proportion of dumbbell tumors was 64% and was similar to the previous published case series. Tumor location within the spinal canal was classified as lateral, anterolateral, or posterolateral, if a clear anterior or posterior extension across the midline was present. C2 schwannomas, more often originated from the sensory root, were predominantly located posterolaterally (60% of C2 cases), while C1 schwannomas, always originating from the motor root, had, in the majority of cases, lateral location.

Clinical presentation and postoperative outcome

Cervical or occipital pain was the more frequent clinical finding, occurring in 57% of cases, resulting similar to other case series.[7,16] Myelopathic symptoms occurred, presenting as motor deficit in 36% of the cases and sensory deficit, in the upper (36% of the cases) and in the lower extremities (21% of the cases). The mean preoperative and postoperative KPS were 77.6 ± 14.6 and 95.3 ± 6.2, respectively, confirming a significative postoperative clinical improvement.

Gross total resection and recurrence rate

Complete surgical removal is generally considered the gold standard treatment for spinal schwannomas to reduce recurrence risks. The previous surgical series of C1–C2 PNSTs showed higher recurrence rates in cases of subtotal resection,[2,11] and there were no recurrences when GTR rates were higher although associated with increased mortality and morbidity rates.[6,14] The association between GTR and increased morbidity rate was further confirmed by Ryu *et al.*,[27] in a study on cervical dumbbell-shaped schwannomas, in which the authors reported better neurological outcome in cases of STR then GTR. However, contrarily to these findings, GTR was accomplished in all cases in our study, associated with a global postoperative improvement, low rate of postoperative complications, absence of spinal instability, and tumor recurrences. Even if limited by the small number of cases, these results demonstrated that for C1–C2 schwannomas, GRT can be safely accomplished and VA involvement or dumbbell tumor shape did not affect the EOR and surgical outcome [Figures 3 and 4].

Nerve root preservation

Schwannomas are benign slow-growing PNSTs that grow as an exophytic mass originating from a single nerve root or involving the entire nerve root intermingled with the nerve fibers. In the former case, nerve root removal was necessary to accomplish complete tumor resection, especially in case of large-size tumors. Therefore, some authors believe that preservation of nerve root is possible only for small schwannomas located intradurally. Conversely, when tumor mass involves the entire motor or sensory rootlet, or extends beyond the nerve arachnoid sheath, the sacrifice of the entire root is required.[29,30] Moreover, other authors reported that, even when the nerve root involved is highly functional, the sacrifice infrequently produces postoperative deficits, probably

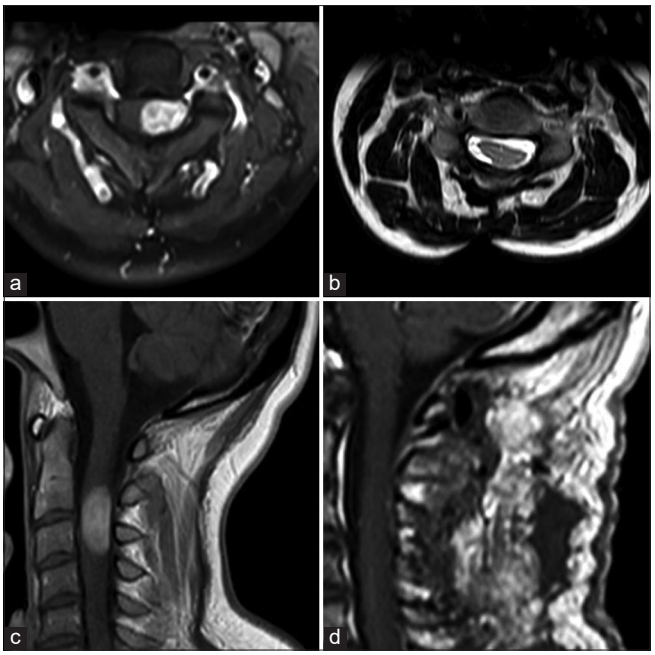

Figure 3: Case 10. Preoperative (a) axial and (c) sagittal contrast-enhanced T1-weighted magnetic resonance images (MRI) showing extradural tumor located posterolaterally to the spinal cord at C2 level on the left side. Postoperative (b) axial and (d) sagittal contrast-enhanced T1-weighted MRI-confirmed gross total resection

Table 4: Surgical approaches according to tumor location

	Tumor location			Total
	Intradural	Extradural	Dumbbell	
Hemilaminectomy	1	1	4	6
Laminectomy	1	0	4	5
Far-lateral approach	0	1	1	2

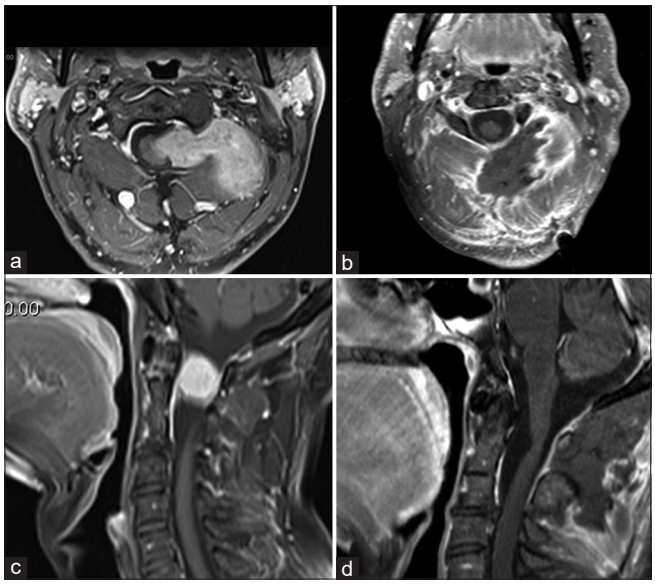

Figure 4: Case 13. Preoperative (a) axial and (c) sagittal contrast-enhanced T1-weighted magnetic resonance images (MRI) showing a dumbbell shaped tumor located posterolaterally at C2 level on the left side. Postoperative (b) axial and (d) sagittal contrast-enhanced T1-weighted MRI-confirmed gross total resection

due to the denervation process operated by the tumor mass.[31] In addition, C1 nerve root lacks sensory fibers and both C1 and C2 nerve roots have numerous distal nerve anastomosis with the cervical plexus, and hence, the sacrifice of these nerve roots produces, rarely, a very mild deficit in a very limited area. Indeed, in our series, small nerve rootlets were routinely sacrificed, but the entire nerve root was completely resected only in one case of a C1 tumor, without clinical consequences.

Vertebral artery management

In this series, VA resulted to be compressed by the tumor in four cases (30.7%) and encased in two (15.3%). All cases with VA involvement were C2 schwannoma. Several authors discussed surgical management of VA for PNST.[17,26,32] George *et al*. suggested that proximal control of VA should be routinely performed before tumor resection.[17] Contrarily, Goel *et al*.[26] demonstrated, in a series of 60 PNSTs of C2 root, that proximal or distal control of the VA was not necessary, because the dural sheath covering the tumor mass separates the VA and the venous plexuses from the tumor itself, and, when dissection is performed within this plane, the risk of VA injury is very low. More recently, Wang *et al*. confirmed this surgical strategy in a series of C2 schwannomas in which they described the venous relationships of dumbbell PNSTs located at C2.[32] However, these studies did not include any cases of encased VA. Tumor encasement of VA implies the invasion of both dural sheet and venus plexus and the remaining of the sole periosteal sheath as VA coverage. Therefore, in those cases, the technique suggested by George is probably more suitable. In fact, in our series, VA encasement was treated performing proximal VA control and extraperiosteal dissection of VA, thus stripping the periosteal and venous sheaths. In the rest of the cases, even when the VA was compressed, tumor dissection was performed in the dural plane between tumor and VA. Finally, no VA injury occurred in the series, and we suggest that proximal artery control is unnecessary for dumbbell cervical schwannomas sparing or only compressing VA, in which an optimal dural dissection plane to perform tumor resection. On the other hand, when VA is encased, surgical dissection has to be conducted to obtain a proximal arterial control and an extraperiosteal dissection of the VA.

Instability

Spinal instability was not observed in our study and previous case series of cervical schwannomas reported the same findings.[10,27,30] However, the loss of bone integrity after cervical laminectomy, the skeletonization of paravertebral muscles of C2 region, and the partial resection of the occipital condyle during far-lateral approach constitute a possible risk of delayed instability.

Therefore, preventing the incidence of spinal instability is recommended when possible. In our series, we preferred to perform a minimally invasive hemilaminectomy, to minimize bone removal, as previously demonstrated by other groups.[33-35] Nonetheless, no conclusions regarding the risk or prevention of delayed instability can be made from this study and from the available literature.

CONCLUSIONS

C1–C2 schwannomas are rare tumors that harbor specific anatomical features that make the surgical resection very challenging. The results of this cumulative series demonstrates that despite the complex surgical managment, complete resection can be accomplished with excellent outcome and very low rate of complications and recurrences.

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Conflicts of interest

There are no conflicts of interest.

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