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Lateral intra-extraspinal cervical schwannomas: Partial versus total resection

Research Article

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Abstract: The study involved 27 lateral cervical schwannomas surgically treated in the last 40 years utilizing either partial or total resection. The study aims to compare the results of total and partial resection of spinal lateral schwannoma and to assess the values of partial resection of large lateral cervical schwannomas. We performed a retrospective analysis of 27 patients operated for lateral cervical schwannomas in the period 1971-2010, out of which 11 had partial resection. The average follow-up of the patients was 136.4 months, comparing pre- and postoperative clinical picture, time to relapse, dynamics of yearly growth of schwannomas, and the need for re-resection. The average period of preoperative complaint in lateral cervical schwannomas was 19.6 months. Unilateral cervico-brachialgia as a manifestation of radiculopathy was observed in 85% of LCSs, and, was,as a rule the first symptom of the disease. Surgical treatment of 27 patients with LCSs resulted in recovery or improvement of complaints in 80% of cases.The treatment effect did not depend on the degree of resection. Reoperation was done, on average, 16.3 years after partial resection. The residues of lateral cervical schwannomas stopped growing 15 years after partial resection and underwent cystic degeneration. Partial resection of lateral cervical schwannomas is an excellent alternative to total resection, not only for the elderly or in cases of increased surgical risk, but also as a method of choice since it is more readily available to a larger number of surgeons and associated with low percentages of surgically relevant relapses and high percentages of improvement.

Keywords: *Lateral cervical schwannoma • Partial resection • Growth arrest • Cystic degeneration*

1. Introduction

Schwannoma or neurilemmoma is a benign tumor originating from the Schwann cells, i.e. the sheath of the peripheral motor, sensory, sympathetic, or cranial nerves. It should be differentiated from neurofibroma, which also originates from the nerve sheath Schwann cells, but contains fibroblasts in almost equal numbers [1]. According to some authors, 25-45% of all extracranial schwannomas are situated in the craniocervical junction or the cervical region [2]. Cervical schwannomas are divided by Daly and Roesler into lateral and medial ones depending on the distance from midline and the nerve of origin [3]. Lateral cervical schwannomas (LCS) originate from the sheath of cutaneous or muscle branches of the cervical or brachial plexus, while medial cervical schwannomas arise from the sheaths of the last 4 cranial nerves, including the vagus nerve, or from the sympathetic fibers of the cervical region [4]. The treatment of schwannoma involves microdissection and complete tumor removal, with mandatory intraoperative sacrifice of the nerve root in 50-60% of

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Table 1. Pre and post-operative statistics in 27 lateral cervical schwannoma patients.

	Characteristics	Lat. cerv. schwanoma No.of cases (%)	Partial resection No.of cases (%)(f/m)	Total resection No.of cases (%)(f/m)
1.	Sex (m/f)	27 (13/14) (100.0)	11 (5/6) (100.0)	16 (8/8) (100.0)
2.	Mean age of patients (in year)	51.2	57.6	47.8
3.	LCS position and type			
	-C2-C3	3 (11.11)	1 (A+B+C)	2 (A+B+C)
	-C3-C4	3 (11.11)	2 (A+B+C)	1 (B+C)
	-C4-C5	4 (14,81)	1 (A+B+C),1(B+C)	2 (A+B+C)
	-C5-C6	6 (22.22)	2 (A+B+C)	3 (A+B+C),1 (B+C)
	-C6-C7	7 (25.93)	1 (A+B+C),1(B+C)	5 (A+B+C)
	-C7-T1	4 (14,81)	1 (A+B+C),1(B+C)	1 (A+B+C),1 (B+C)
4.	Schwanoma type A+B+C	21 (77.77)	8 (78.73)	13 (81.93)
	Schwanoma type B+C	6 (22.23)	3 (21.27)	3 (18.07)
5.	Pre-operative symptoms:	11 (40.74)	4 (36.36)	7 (43.75)
	Local back pain	23 (85.18)	9 (81.81)	14 (87.50)
	Radiculopathy	7 (25.92)	2 (18.18)	5 (31.25)
	Myelopathy	6 (22.22)	2 (18.18)	4 (24.00)
	Motor weaknes	4 (14.81)	1 (9.09)	3 (18.75)
	Voiding difficulty			
6.	Average pre-operative duration of the symptoms (in months)	19.6	18.3	21.7
7.	Multiplicity	1	1 (9.09)	0
8.	Recurrence/tumor progression	11 (40.74)	10 (90.90)	1(6.25)
9.	Reoperation	3 (11.11)	2 (18.18)	1(6.25)

cases [5]. In almost 30% of surgically treated cases, this results in permanent neurologic deficiency; therefore, Gore and Katz have suggested partial resection for schwannoma, where we intentionally leave a small portion of the tumor on a nerve root [6,7]. Unfortunately, in 55% of partially resected schwannomas, a surgically relevant relapse occurs, requiring re-resection [8]. Schwannoma relapses can be accompanied by malignancy in 4% of the cases [9].

1.1. Aim of the study

In the group of surgically treated lateral cervical schwannomas (LCS), monitored for years, the study aimed to compare the results of total and partial resection of spinal lateral schwannomas through the pre- and postoperative periods, including neck mobility, neurologic weakness, relapse dynamics, and possible presence of malignant degeneration of partially resected LCSs; and to validate the approach of partial resection for large LCSs.

2. Method of Work

The paper retrospectively analyzed 27 patients with LCS (14 men, 13 women) surgically managed at the Clinic of Neurosurgery, Clinical Centre Niš, from January 1971 to October 2010, out of which 10 with partial schwannoma resection. The following elements were compared: pre- and postoperative clinical picture, time to relapse, schwannoma yearly growth dynamics, and need for re-resection. The position and size of schwannoma was determined using the diagnostic methodology availble to the field at the time of assessment, that is to sayspinal myelography (SM) – lumbar ascendent and suboccipital descendent myelography until 1984 (9 patients), native and postcontrast computerized tomography (CT) from 1985 to 1995. (11 patients), and magnetic resonance imaging (MRI) from 1996 to 2010 (7 patients). In cases of myelographic diagnostics, due to the lack of information considering the tumor extension and its relations to the surrounding neurovascular structures, the authors prefered partial resection. Pathohistological tumor verification with the H&E technique was performed for all patients, and from 2004 onwards with S-100 protein staining as well. Surgically treated schwannomas were monitored for 136.4 months on the average (range, 6-480 months).

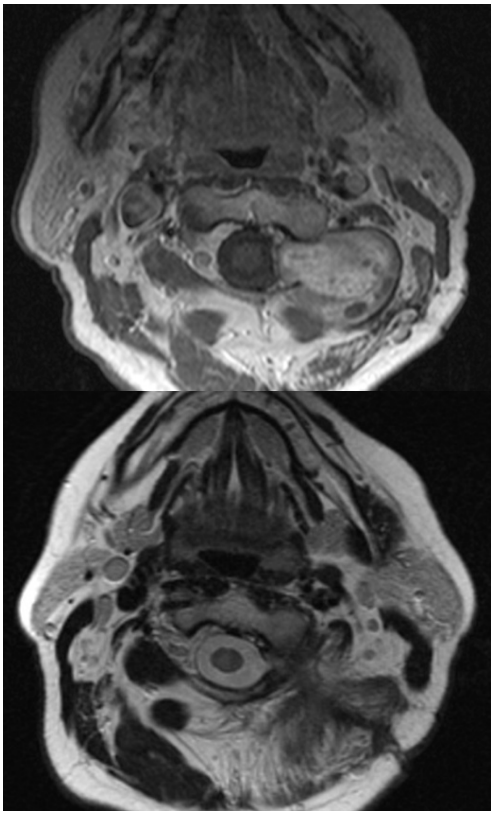
Figure 1. Preoperative sagital T2-weighted magnetic resonance imaging shows an intra-extradural extramedullar dumbbell schwannoma (type A+B+C) on the C6-C7 level in a 53 year-old man.

3. Results

During the period of four decades, 27 patients with LCSs were treated in our institution, of which 13 were men and 14 women (Table 1). The surgeries were performed by two neurosurgeons. The first was in charge until 1994 and the other one from 1994 forward. The tumors were divided into 3 types depending on their anatomical location and relationship with the dura as identified during surgery [10] (Table 1). Type A tumors were those that were located within, or extended into the spinal dural tube.Type B tumors were those that were located posterior to the lateral masses , as an interdural tumor because it was located within the dural tube of the ganglion . Type C tumors were those tumors that extended outside the limits of the spine and extended into paraspinal spaces (Figure 1, 2).

In 11 cases (40.74%), partial schwannoma resection was utilized, involving single or double laminectomy with resection of the spinal root and resection of the schwannoma portion situated within the spinal canal and intervertebral foramen. In seven of these cases, the diagnosis made by lumbar and suboccipital myelography were , resulting in a lack of information about the extension of schwanomma into the prevertebral compartment and its relation to the vertebral or carotid artery, jugular vein or vagus nerve. Absence of mobility of the remaining cervical schwanomma in prevertebral compartment during the surgery as a sign of its extensivity and to the vascular structures was

Figure 2. Pre and postoperative axial magnetic resonance imaging in a 51 year-old woman with total resection of C2-C3 lateral cervical schwannoma type B+C.



the decision criterion for abandoning radical resection with posterior approach. When the extension of the schwannoma in the prevertebral compartment and the relation to neurovascular structures was known, the authors prefered radical resection in two separate surgeries: initially posterior followed by an anterior approach. Thermocoagulation of extraforaminal tumor portion was done to prevent bleeding and stimulate fibrosis within the intervertebral foramen, in order to prevent tumor extension into the spinal canal. Among the schwannomas treated this way were 2 patients (7.4%) with *dumbbell* schwannomas (Figure 1).

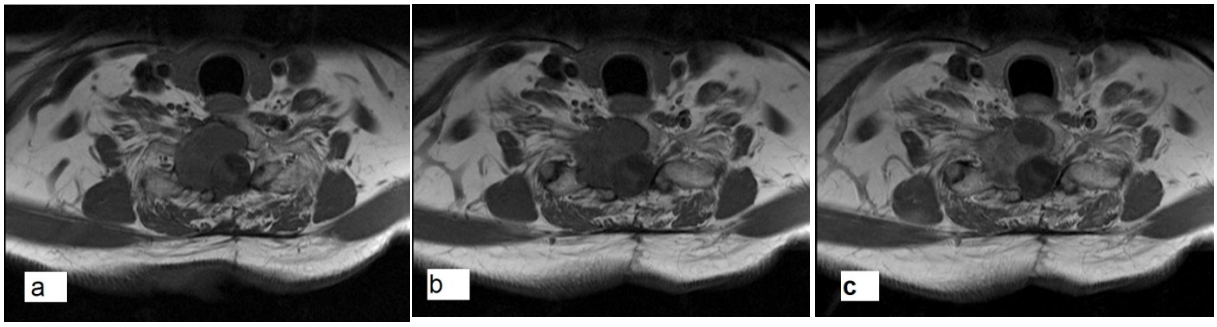
In four patients, the radical resection of cervical schwanomma was a result of patients' resigning from the planned second phase of surgery due to advanced age (over 70yrs: two patients), neurological improvement achieved by the first phase of surgery (1 patient) or the need for complex instrumental fixation (1 patient). Somatosensoric evoked potentials were intraoperatively applied in operating the last 9 patient (in 3 patients with partial and 6 patients with total resection).

In the remaining 16 patients (59.26%) with LCSs, total tumor resection was done, with half involving both the anterior and posterior approach. The patients were

Table 2. Postoperative clinical results in 27 surgically treated patients with lateral cervical schwannomas.

Clinical results	Number of Cases (%)					
	Partial resection		Total resection		Σ	
	11	(100,0)	16	(100,0)	27	(100,0)
Recovered	6	(54,55)	10	(62,50)	16	(59,26)
Improved	3	(27,27)	3	(18,75)	6	(22,22)
Stable	2	(18,18)	1	(6,25)	3	(11,12)
Worsened	0		2	(12,50)	2	(7,40)
Dead	0		0		0	

Figure 3. Lateral cervical schwannoma in a patient 12 (a), 15 (b), and 18 (c) years after operation. Axial magnetic resonance demonstrated a contact between the schwannoma and the dural sac, without its compression and deformation. There was no visible tumor growth. The phase of tumor growth stagnation was replaced by the phase of cystic degeneration.



51.2 years old on average; partial resection was done in the group of patients whose average age was 57.6 years, compared to the group with total resection where patients were almost a decade younger.

LCSs had transdural extension in 77% (Table 1), which required subdural revision. Although the restitution of dural barrier carried the risk of cerebrospinal fistula (CSF), liquorrhea was observed in only 2 of 27 cases (7.40%), with operative revision performed in just one case, in which spontaneous stoppage of liquor leakage could not be observed even after 72 hours postoperatively.

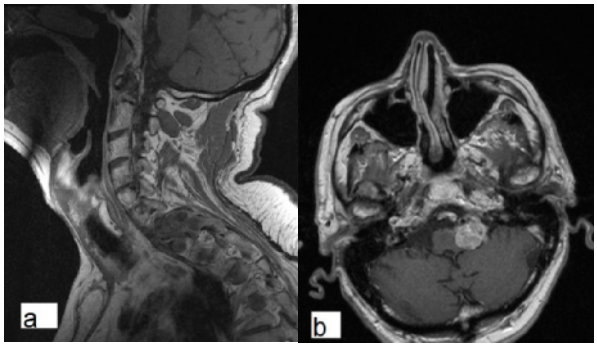
Unilateral cervicobrachialgia, as a manifestation of radiculopathy, was observed in 85% of LCSs, as the first disease symptom as a rule. In 3 out of 11 patients with local painful cervical syndrome, the local pain was the first symptom of their LCS. Everyday neck pain resulted in a more rapid undertaking of diagnostic procedures compared to cervicobrachialgia. The average period of preoperative complaints produced by LCSs was 19.6 months (23.6 months in patients with cervicobrachialgia, and only 7.4 months in those with local pain in their cervical spine; $p<0.01$).

Total resection was performed in 16 (59.26%) out of all LCSs (Figure 2), with two-phase surgery in half of the treated, utilizing firstly the posterior approach and within 2 weeks, anterior approach as well. In 6 patients (22.2%) some of the techniques of stabilization and fixation of the cervical spine were utilized (autografts from the crista iliaca or tibia, anterior titanium plate, and/or posterior fixation through the lateral masses from C3 to C7 in one case). In the remaining cases, the patients wore Philadelphia-type neck immobilizers (collars) for 8 weeks.

Comparing the complaints and events before and after surgery, we observed that surgery resulted in recovery or improvement of symptoms in 80% out of 27 patients with LCS, with the treatment effects being identical in both LCS subgroups (Table 2). At the same time, in 2 patients (7.40%) with total LCS resection the situation deteriorated (reduced neck mobility in anteflexion and lateral rotation by over 10 degrees, occurrence of vertiginous crises and Horner's syndrome). These were patients with extensive LCSs, compressing their prevertebral portion the vertebral artery preoperatively, simultaneously leading to the osteoresorption of more than two thirds of adjacent vertebral bodies (C4 and C5), so that the surgical treatment warranted both approaches and bone graft fixation from C3 to C6.

Surveillance of the dynamics of growth/enlargement of LCS residua/relapses revealed that the average postoperative volume of the residual tumor was 2.418 mm^3 in patients treated with partial resection for LCS using automatic determination of tumor volume on MRI or using the tumor diameter in three planes and mathematical calculation by Pinski

Figure 4. Eighty-two-year-old man with LCS on the right side of C7-T1 level (a) and left side of cranio-cervical junction (b). Magnetic resonance imaging shows cystic schwannoma degeneration in both views.



(**V = a** x **b** x **c** x **0.523**) [11]. Tumor volume determination in several years after surgery revealed an increase of the volume of residual tumor LCS up to ¼ of the volume a year, up to 8th year, after which the dynamics of volume enlargement decreased to 5-10% and completely dissapeared 15 years after surgery (6 patients), when it reached the average of 64.9 cm^3. After that period, the LCS volume slightly decreased, with more intense cystic degeneration and discrete involution (Figure 3). In the period mentioned, temporary aggravation of the clinical picture was observed in 3 out of 11 patients with partial LCS resection (27.2%), and after reoperation with posterior approach and decompression in the spinal canal and intraforaminal space, the patients continued to live without any complaints. Reoperation was done after 16.3 years on the average (range, 11-22 years).

The only patient with a spinal cervical schwannoma and schwannoma in the craniocervical junction (a male aged 82 years, still completely functional and asymptomatic 18 years after partial resection for LCS of C7-T1), who had not agreed to the surgical treatment of schwannoma demonstrated by magnetic resonance in the craniocervical junction, did experience tumor growth stagnation and cystic degeneration which coincided completely with the occurrence in LCS (Figure 4).

4. Discussion

According to some authors, schwannomas displace the nerve from the sheath from which they arise during their growth, so that the nerve is situated at the tumor periphery. Tumor specimen staining with silver has confirmed that the axons are not surrounded by tumor tissue but instead are bound to its capsule [12]. The information theoretically opens the possibility of sparing the nerve from the sheath from which the tumor has arisen by way of careful microscopic tumor dissection. Unfortunately, in surgery of schwannoma, identification of such fibers within the schwannoma capsule is not feasible; the above information carries no practical relevance – in 5 out of 6 schwannomas the nerve has to be sacrificed [13]. The information that partial schwannoma resection results in 29% of functional decay of the involved nerve and that in 4% of residual schwannomas malignant alteration occurs, should compel us to prefer total over partial resection [14,15]. On the other hand, unilateral or bilateral facetectomy which has to be done with total resection, necessarily creates the conditions for angular kyphosis of the spine, and schwannoma adherency to the dural sac, as well as the dislocation of vertebral artery, increases the risk of complications during tumor removal and fixation through the lateral masses in the posterior surgical approach as the first step in two-phase surgery [16]. If we also take into account the fact that there are almost no studies monitoring the residues of schwannoma over several decades, the advantage of total, over partial, resection of spinal CLS could not be decided upon in an evidence-based way [17].

The average duration of complaints before diagnosis and operation was 19.6 months in this study, confirming the results obtained by other authors about delayed recognition of cervical schwannomas due to their slow growth, symptomless course, and non-specific symptoms (cervicobrachialgia in 85% of cases in this paper), being frequently mistaken for cervical spondilosis or discopathy.

The comparison of clinical status before and after surgery revealed a significantly better outcome from partial resection (80%), which was not inferior to that observed for the approach with total resection. The result was even better, since there were no additional neurogenic lesions or complications compared to the group with total resection done for LCS (with complications in 7.40% of cases). Decompression of nerve structures within the spinal canal, in the situation of a slowly growing schwannoma with clinically relevant relapse after 11 years or longer, represents a long-term benefit for the patient. Therefore, partial resection of LCS, with tumor removal from the spinal canal and intervertebral foramen, even with sacrificed spinal roots, did not result in a permanently aggravated status.

All residual LCSs demonstrated a tendancy to relapse, but the newly-created tumor mass was distributed asymetrically, with predominant extraforaminal, prevertebral presence. Surveillance of the volume growth of residual schwannomas revealed a gradual, unvaried progression of volume growth in the first 7-8 years of less than 25% of tumor volume a year, with significantly decreased growth after that. On the average, LCSs stop growing after 15 years – a balance between growth and

cystic degeneration of LCSs is then reached. If a LCS has not produced compression of nerve structures in the 15 years after the last operation, the tumor is no longer clinically relevant. Although schwannomas are associated with small cysts from the very beginning of their development due to vacuolar degeneration in Antoni B areas, larger cysts are rarely seen in the period, being, as a rule, the consequence of hemorrhagic degeneration of schwannomas, as confirmed by the finding of hemosiderin within [18]. Histologically, schwannomas consist of compact cellular lesions (Antoni type A tissue) and loose, hypocellular myxoid lesions with microcystic spaces (Antoni type B tissue). Benign schwannoma sometimes displays degenerative changes, such as cyst formation, calcification, hemorrhage, and hyalinization [19]. Macrocystic degeneration was detected in more than 20% of schwannoma diagnosed by aspiration cytology. Smears from schwannomas with cystic degeneration showed scattered round-to-oval cells, accompanied by occasional histiocytes [20]. The authors report cases with macrocystic degeneration of orbital, pancreatic , peripheral nerve sheath and retroperitoneal schwanomas [21-24]. The degeneration is due to central tumor necrosis as the schwannoma grows to a size beyond the capacity of its blood supply in maintaining survival of central areas [25].

Cystic degeneration of LCSs observed in this study 15 years after surgery was not accompanied by hemosiderin deposits and finally confirmed the arrest of growth and initial schwannoma involution. The fact that cystic degeneration was observed simultaneously in another, smaller schwannoma in the same patient, suggested a temporally programmed, genetically determined cystic degeneration, but the confirmation of the proposition would require the study of a larger series of patients.

5. Conclusion

Partial intraspinal resection of lateral cervical schwannomas is associated with clinical improvement in over 80% of surgically treated patients, and is not inferior at all to total resection.

In LCS residues, a growth of tumor volume of 25% a year has been observed in the first several years, with subsequent arrest of growth and cystic degeneration 15 years after partial resection.

In one fifth of partially resected LCSs, re-operation is required to treat surgically relevant relapses. Surgical treatment of LCS relapses has been applied 10 or more years after partial LCS resection.

Partial resection of LCSs is an excellent alternative to total resection not only for the elderly or patients at increased surgical risk, but it is also a method of choice since it is available to a significant number of surgeons and associated with low rates of surgically relevant relapses and high rates of clinical improvement.

References

[1] Abell MR, Hart WR, Olson JR. Tumors of the peripheral nervous system. Hum Pathol 1970;1:503-51

[2] Leverstein H, Castelijns JA, Snow GB. The value of magnetic resonance imaging in the differential diagnosis of parapharyngeal space tumors.Clin Otolaryngol 1995;20:428-33

[3] Daly JF, Roesler HK. Neurilemmoma of the cervical sympathetic chain.Arch Otolaryngol 1963;77:262-6

[4] Som P, Biller H, Lawson W. Tumors of the parapharyngeal space. Preoperative evaluation, diagnosis and surgical approaches. Ann OtolRhinol Laryngol 1981;90 (suppl 80):3-15

[5] Valentino J, Boggess MA, Ellis JL, Hester TO, Jones RO. Expected neurologic outcomes for surgical treatment of cervical neurilemmomas. Laryngoscope 1998;108:1009-13

[6] Gore DO, Randow R, Harnford JM. Parapharyngeal neurilemmoma. Surg Gynecol Obstet 1956;103:193-201

[7] Katz AD, Passy V, Kaplan L. Neurogenous neoplasms of major nerves of face and neck. Arch Surg 1971;103:51-6

[8] Seppala MT, Haltia MJ, Sankila RJ, Jaaskelainen JE, Heiskanen O. Longterm outcome after removal of spinal schwannoma: a clinicopathological study of 187 cases. J Neurosurg. 1995;83:621-6

[9] Chang SC, Schi YM. Neurilemmoma of the vagus nerve. A case report and brief literature review. Laryngoscope 1984;94:946-9

[10] Goel A, Muzumdar D, Nadkarni T, Desai K, Dange N, Chagla A. Retrospective analysis of peripheral nerve sheath tumors of the second cervical nerve root in 60 surgically treated patients.J Neurosurgery Spine 2008; 8:129-134

[11] Pinski J, Halmos G, Shirahige Y, Vittliff L.J, Shally V.A. Inhibition of growth of the human malignant glioma cell line (U87MG) by the steroid hormone antagonist RU486. J Clin Endocrinol metab 1993;77,1388-1392

[12] De Girolami E, Frosch M, Antonhy D. The central nervous system.In: Cotran R, Kumar V, Robbins S, eds. Robbins Pathologic Basis of Disease.

Philadelphia: W.B. Saunders; 1994. p. 1351-2
[13] Valentino J, Boggess MA, Ellis JL, Hester TO, Jones RO. Expected neurologic outcomes for surgical treatment of cervical neurilemmomas. Laryngoscope 1998;108:1009-13
[14] Seppala MT, Haltia MJ, Sankila RJ, Jaaskelainen JE, Heiskanen O.Longterm outcome after removal of spinal schwannoma: a clinicopathological study of 187 cases. J Neurosurg. 1995;83:621-6
[15] Chang SC, Schi YM. Neurilemmoma of the vagus nerve. A case report and brief literature review. Laryngoscope 1984;94:946-9
[16] Lonstein JE : Post-laminectomy kyphosis. Clin Orthop Relat Res :93-100, 1977
[17] Jinnai T, Koyama T : Clinical characteristics of spinal nerve sheath tumors : analysis of 149 cases. Neurosurgery 2005;56 : 510-515
[18] Enzinger FM, Weiss SW. Soft Tissue Tumors, 2 nd Ed. Toronto: Mosby; 1988, 725-35
[19] Ogose A, Hotta T, Sato S, Takano R, Higuchi T. Presacral Schwannoma With Purely Cystic Form; Spine 2001; 26 (16) : 1817-1819
[20] Domanski HA, Akerman M, Engellau J, Gustafson P, Mertens F, Rydholm A. Fine-needle aspiration of neurilemoma (schwannoma). A clinicocytopathologic study of 116 patients. Diagn Cytopathol 2006; 34:403-12
[21] Subramanian N, Rambhatia S, Mahesh L, Menon SV, Krishnakumar S, Biswas J, Noronha OV. Cystic Schwannoma of the Orbit—A Case Series. Orbit. 2005, 24, (2) , 125-129
[22] Hirabayashi K, Yasuda M, Umemura S, Itoh H, Itoh J, Yazawa N, Imaizumi T, Yoshiyuki OR. JOP. J Pancreas (Online) 2008; 9(2):203-208
[23] Pilavaki M , Chourmouzi D , Kiziridou A , Skordalaki A, Zarampoukas T, Drevelengas A. Imaging of peripheral nerve sheath tumors with pathologic correlation: Pictorial review. European Journal of Radiology 2004; 52(3):229-239
[24] Cury J, Ferreira RC, Srougi M. Retroperitoneal schwannoma: Case series and literature review. CLINICS 2007;62(3):359-62
[25] Donnal JF, Backer ME, Mahony MS, Leight GS. Benign retroperitoneal schwannoma. Urology 1988; 31: 332–4