

Surgical outcome of spinal schwannoma and neurofibroma

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SUMMARY

OBJECTIVE: The aim of this study was to evaluate the outcome and surgical complications in patients with spinal schwannoma or neurofibroma surgically treated at the Hospital das Clínicas of the State University of Campinas.

METHODS: This was a retrospective cohort study, using medical records of patients operated between 2011 and 2021. The sample distribution was verified using the Kolmogorov-Smirnov test. The dynamics between qualitative variables were assessed using Fisher's exact test. We used means analysis to assess patient improvement based on Frankel scores.

RESULTS: A total of 16 patients were evaluated, of whom 56.25% (9) were men and 43.75% (7) were women. There were 13 (81.25%) patients with schwannomas and 3 (18.75%) with neurofibromas. Patients with deficits had neurological improvement, such as walking or with at least Frankel D or E after surgery. Laminectomy, performed in 8 patients (50%), and laminoplasty, used in 9 patients (56.25%), were the main techniques.

CONCLUSION: The surgical approach was proved to be an effective and safe alternative to the treatment of these tumors, with neurological improvement and minor surgical complications.

KEYWORDS: Quality of life. Spinal cord compression. Neurofibroma.

INTRODUCTION

Spinal tumors can be classified according to their relationship with the spinal cord in intradural and extramedullary, intradural and intramedullary, or extradural¹. The intradural and extramedullary tumors correspond to about 40% of the cases of spinal tumors, and the main histological representatives of this group are the schwannomas and neurofibromas, both of these tumors originated from the peripheral nerve sheath, besides meningiomas¹⁻⁴. Patients with intradural and extramedullary tumors generally present with axial pain and neurological symptoms due to progressive compression of the spinal cord, and the main therapeutic modality is surgical resection, given the high efficacy and safety of the method⁵⁻⁹. The resection when performed with a wide exposure of the tumor and with the proper surgical precaution proves to be curative for patients. Surgery has immediate and lasting efficacy, although, in isolated cases, other treatment modalities may play a role, such as radiosurgery, in selected situations⁵.

Nerve sheath tumors, schwannomas and neurofibromas, are the most common forms of intradural and extramedullary tumors. Schwannomas are the most common, followed

by neurofibromas and ganglioneuromas^{1,10}. About 35-45% of patients affected by these tumors suffer from neurofibromatosis (NF), with NF1 related to neurofibromas and NF2 related to schwannomas¹. We emphasize that both can still be found singly or sporadically, especially schwannoma, unrelated to NF. Schwannomas are neoplasms derived from Schwann cells, and most of these tumors originate from the dorsal roots and less frequently originate from the ventral roots, where surgical access is more difficult, as, in general, the standard exposure of the spine is performed by approaches to the posterior midline of the spine²⁻⁵. These tumors are usually associated with radicular pain as well as sensory deficits. Neurofibromas, on the contrary, are commonly involved with the cervical spine, which can make surgical resection difficult, especially when plexiform and involving multiple segments, typical of type 1 NF³. Thus, it is clear the importance of evaluating the treatment of these tumors, as, despite being generally benign, they can cause severe neurological sequelae and great impact on quality of life due to spinal cord compression. The identification and treatment of these neoplasms provide great gain for the patient's quality of life, and the treatment is generally

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curative. Considering the lack of national studies evaluating surgery in our country, the aim of this study was to evaluate the role of surgical treatment in the management of these tumors in a Brazilian tertiary clinical hospital.

METHODS

Patient selection

We performed a retrospective cohort study. Data collection was carried out at the Clinical Hospital of the State University of Campinas. It included all patients with spinal schwannoma or neurofibroma operated on by the neurosurgery team between 2011 and 2021 who met the inclusion criteria. The inclusion criteria were patients with spinal schwannoma or neurofibroma who were surgically treated at the institution, performed by the same surgeon (AFJ). The exclusion criteria were patients who had not undergone surgical treatment and whose data were either missing or not available for analysis. The project was submitted and approved by the Medical Ethics Committee of UNICAMP (CAAE: 48731721.0.0000.5404). Importantly, the confidentiality of these patients was maintained, and their identities were preserved. This is a retrospective study, so there is no risk to the patients involved in this study.

Data collection and definition of some indicators

The clinical data of patients were retrospectively collected. Neurological status was evaluated before and after surgery using the Frankel classification for spinal cord injury. Histological data, as well as information about surgical treatment were collected. The patients included in the study were followed up until the last follow-up visit before data analysis. The medical records were requested and evaluated, and after applying the inclusion and exclusion criteria, the patients included in the study were added to an Excel spreadsheet together with the information of interest to the project. Using the data obtained, a descriptive statistical analysis was performed in order to answer the objectives proposed by the study. The patient characteristics that were selected for the study were length of hospitalization, occurrence of complications, and postoperative functions, and the results were analyzed.

Statistical analysis

After data collection, the results were analyzed. The data obtained were then processed with Excel. To verify the normality of the quantitative variables, the Kolmogorov-Smirnov tests were applied for the age of the patients, whereas the qualitative variables will be presented in absolute and relative values. The evaluation of the association between variables was done using the

chi-square test. Furthermore, mean improvement analysis was performed to evaluate the improvement of neurological status between Frankel scores before and after surgery.

RESULTS

The analysis included 16 patients after applying the inclusion and exclusion criteria; 56.25% (9) were men and 43.75% (7) were women, so it was not possible to identify a preference for a specific gender. The mean age of the population was 44 years, with 41.5 years for males and 47.3 years for females. We used the Kolmogorov-Smirnov test (age analysis), aiming to verify if there is a normal distribution of the data, and from this, we could identify some biases. The overall age range was 20-71 years, 25-67 years in women and 20-71 years in men. Regarding tumors, there were 13 patients with schwannomas (81.25%) and 3 with neurofibromas (18.75%). All patients who were affected by neurofibromas had the diagnosis of type 1 NF (100%). We did not observe recurrences in our sample during the study follow-up. In one patient, it was necessary to divide the surgical procedure into four surgical steps—two for tumor removal and two for deformity correction. This strategy was used due to the high complexity of the procedure, in which it was necessary to perform an intra- and extradural resection with the release of the vertebral artery. One year after the removal of the tumor, due to an iatrogenic kyphosis, it was necessary to perform a cervical arthrodesis between C3 and C6 in this patient.

The tumors presented an uneven distribution as to location, 7 (43.75%) were ventrally located, 6 (37.5%) were lateral to the spinal cord, and 5 (31.25%) were dorsal; among all the tumors, one had ventral and dorsal location simultaneously and another had lateral and dorsal location. No correlation was found between the location of the tumors and the clinical recovery of the patient.

Regarding the correlation of tumor location with the main complaint, we noticed a tendency for more cranial tumors to have motor deficits as the main complaint, as we can see in Figure 1. To test this correlation, Fisher's test was applied to the sample. The p-value for our sample was 0.023601, which means that, considering that the distribution is random, the chance of this scenario to occur is 2.36%. Considering an alpha of 0.05, we have to reject the null hypothesis, which predicted an absence of a relationship between the location and the main complaint of the patient, so there is an association between the location of the tumor and the patient's main complaint. The main symptom found in the patients was very severe pain, and the information about irradiation was compromised because many medical records did not describe whether or not there was irradiation of pain.

The analysis of the average improvement was made, and each patient had an average increase of 0.8125 on the Frankel scale, a mode of increase of 1 on the scale, with most moving from D to E. When analyzing the data, there was a trend of improvement on the Frankel scale. In Table 1, a comparison between Frankel score in the preoperative and postoperative stages is presented, indicating that the distribution is altered through the surgical intervention. The mode was that the patients had an improvement of 1°, with the majority moving to grade E on the scale, as we can see by comparing Figures 2 and 3. It was identified that 14 (87.5%) of patients had spinal cord compression by the tumor, of which two had a worse score on the Frankel scale, being classified as B. Interestingly, even patients with severe spinal cord compression (Frankel B) showed significant improvement with surgery, and 11 (68.75%) patients had an improvement of at least 1° in the Frankel score.

The main techniques used were laminectomy and laminoplasty. Laminoplasty was used as one of the surgical techniques of approach in 9 (56.25%) patients, whereas laminectomy was done in 8 (50%) patients and 1 patient underwent both techniques. Laminectomy had to be associated with other techniques in 7 (87.5%) patients, such as facetectomy, concomitant arthrodesis, and duroplasty, among others. Arthrodesis was performed in

3 patients (18.75%) together with laminectomy, and 1 patient had to undergo circumferential arthrodesis 1 year after tumor removal due to iatrogenic kyphosis. Laminoplasty was associated with two other procedures (i.e., laminectomy and flavectomy) in 2 (22.22%) of the patients. In figure 4 we can see an example of the surgery procedure with demonstrative images.

Two patients had to undergo drainage of the thorax due to postoperative bleeding of intrathoracic tumor removal. There were also two cases of thoracic bleedings in which the patients required a chest drain during the initial postoperative period. One of these patients, during return visits, complained of chest pain, which was controlled with medication. One of the patients operated on for neurofibroma, during the return visit, complained of leg pain; the pain was coming from new neurofibromas in the leg region. The patient had the diagnosis of NF1. There was a case of surgical wound infection, treated with ciprofloxacin and with resolution without the need for further surgery. There was also one patient with micturition disorder during the post-surgery period who was referred to the urology service to investigate the origin of the problem. Regarding pain, two patients remained with chronic pain in the postoperative period and required medication for pain of neuropathic origin. Thus, despite having complications (5 of

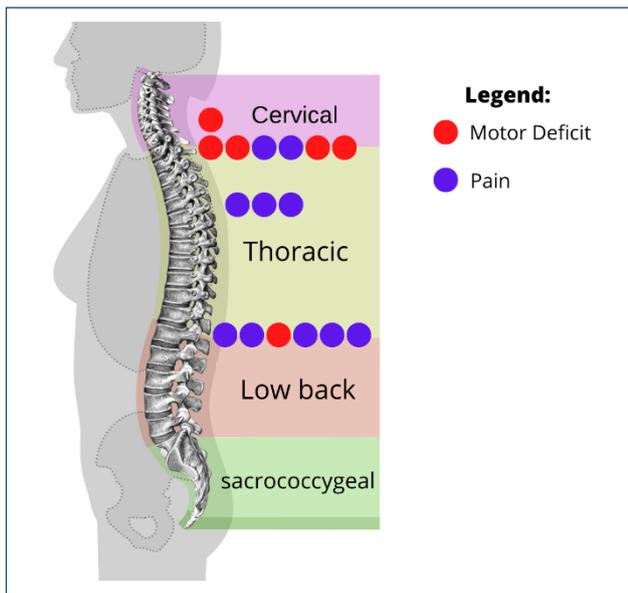


Figure 1. Presentation of symptoms.

Table 1. Comparative improvement of Frankel grade before surgery and postoperative at the final follow-up.

	A	B	C	D	E
Preoperative Frankel	0	2	0	12	2
Postoperative Frankel	0	0	0	5	11

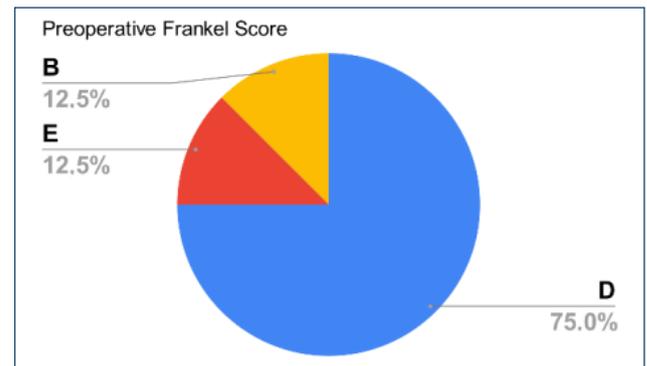


Figure 2. Preoperative Frankel score.

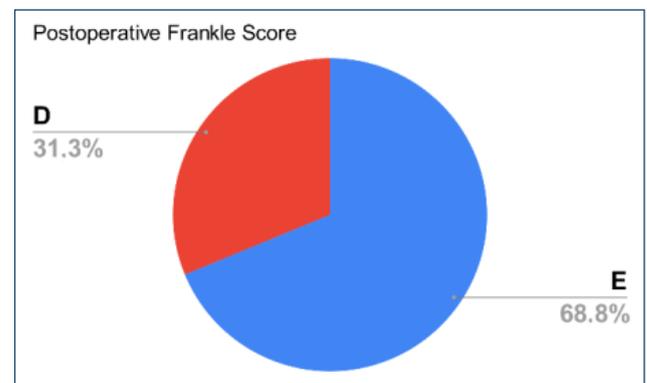


Figure 3. Postoperative Frankel score at the last follow-up.

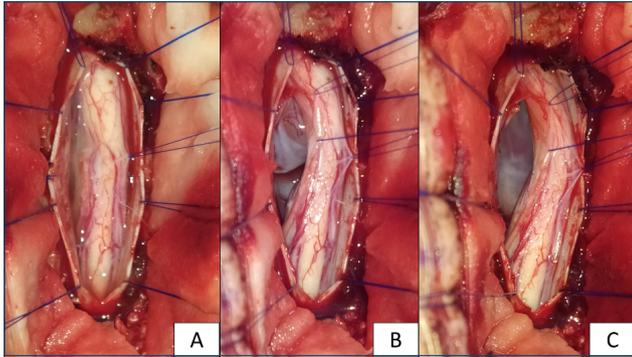


Figure 4. Patient with thoracic ventral schwannoma. (A) Initial exposure after dural opening. The tumor cannot be seen initially. (B) After sutures in the dentate ligament, the spinal cord is rotated to the contralateral side, with good tumor visualization. (C) Surgical image after total tumor removal without any additional injury to the spinal cord.

the 16 patients) such as bleeding, infection, fistula, difficulty to urinate, and chronic pain, no patient had serious sequelae from the surgery. Thus, joining this to the benefits that were brought to the patients, we classified the surgical approach as an effective and safe alternative for intradural tumors.

DISCUSSION

In this study, we present a series of 16 patients who underwent surgical treatment of tumors derived from the neural sheath of peripheral nerves in a single center. There was a significant neurological improvement in all patients with deficits, and at the end of the study, all were walking and with at least Frankel D (preserved strength in four limbs and able to ambulate). Among those found in the sample, we observed a higher prevalence of schwannomas when compared with neurofibromas, with no gender preference. This is probably due to the high rate of patients with sporadic schwannomas (not associated with NF2) in the studied region. Among the patients with neurofibromas, all had NF1, demonstrating the correlation between the appearance of tumors and this underlying genetic disease. The main symptoms found were motor deficit and pain, and a correlation between tumor height and the main complaint was demonstrated. More cranial tumors have a higher incidence of motor complaints.

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As for the prognosis, it was noticed that, in general, the surgical approach provided clinical improvement to the patients, with a reduction of motor deficit and pain. Most patients were classified as E on the Frankel scale after surgery. Regarding the approach and techniques, we would have to increase the number of patients to identify the prognosis of the patients approached via the ventral approach. The main techniques used were laminectomy and laminoplasty. In two cases, a late reapproach was necessary because of kyphotic deformity after laminectomy and because of residual tumor growth (partial resection).

Limitations

This was a retrospective study with limited statistical power, rather than a prospective cohort study. Besides, the number of patients enrolled in this study was not enough. Anyway, the benefits of surgery were clearly demonstrated.

CONCLUSION

The surgical results were considered good because there were neurological improvements, no major complications, deaths, or neurological worsening in this series. Schwannoma-type tumors not related to NF predominated in our series. No patient had severe neurological sequelae. The surgical treatment at our institution was safe and effective, with acceptable complication rates and few neurological sequelae.

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AUTHORS' CONTRIBUTIONS

JVCT: Data curation. **AFJ:** Formal Analysis, Project administration, Supervision, Writing – review & editing. **ACFF:** Conceptualization, Investigation, Methodology, Software, Validation, Visualization, Writing – original draft. **CMBB:** Writing – original draft. **GBM:** Data curation.

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