Surgical management of sacral schwannomas: a 21-year mayo clinic experience and comparative literature analysis

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Abstract
Introduction Sacral and presacral schwannomas are rare, accounting for a minority of spinal schwannomas. We present our institution’s experience surgically treating spinal schwannomas and compare it to the literature.

Methods Data were collected for 27 patients treated surgically for sacral or presacral schwannoma between 1997 and 2018 at all Mayo Clinic locations and 93 patients in the literature. Kaplan–Meier disease-free survival analysis was conducted. Unpaired two-sample t tests and Fisher’s exact tests assessed statistical significance between groups.

Results Our patients and those in the literature experienced a similar age at diagnosis (49.9 y/o. vs 43.4 y/o., respectively). Most of our patients (59.3%) reported full recovery from symptoms, while a minority reported partial recovery (33.3%) and no recovery (11.1%). A smaller percentage in the literature experienced full recovery (31.9%) and partial recovery (29.8%) but also no recovery (1.1%). Our patients experienced fewer complications (14.8% versus 25.5%). Disease-free survival curves for all patients showed no significant variation in progression by extent of resection of schwannoma (log-rank P = 0.26). No lesion progression was associated with full or partial symptom improvement (p = 0.044), and female patients were more likely to undergo resection via a posterior approach (p = 0.042).

Conclusion Outcomes of patients with sacral or presacral schwannomas vary based on patient demographics, tumor characteristics, symptoms, and surgical treatment. Among the range of symptoms experienced by these patients, the most common is pain. Prognosis improves and overall survival is high when the surgical approach towards sacral schwannomas is prepared and executed appropriately.

Keywords Schwannoma · Sacral · Presacral · Surgical management · Outcome

Introduction
Schwannomas are benign, slow-growing neoplasms of the peripheral nerve sheath, often arising from the dorsal rootlets of the spine. These are the most common tumors of peripheral nerves and present in the spine as intradural, extramedullary tumors. While spinal schwannomas typically present in the thoracic region, 1–5% originate in the sacrum [1]. Schwannomas may develop sporadically or be associated with inheritance. For instance, neurofibromatosis 2 is associated with the development of various benign tumors throughout the central nervous system, spine, and periphery, and up to 75% of patients with schwannomatosis develop spinal schwannomas [2].

The clinical presentation of sacral and presacral schwannomas (tumors contained within the presacral space or extending into it from the sacrum or foramina) may involve local pain and weakness, changes to sensory innervation of the bowel and bladder, or sexual dysfunction. Due to the reality that these tumors may remain asymptomatic for extended periods of time, patients may present with large lesions later discovered in the fourth and fifth decades of life often following the development of neurologic symptoms [3].
Except for patients with a history of neurofibromatosis type 2, sacral schwannomas often follow an indolent course postoperatively with low rates of recurrence or malignant transformation [5]. While total resection has the potential to relieve symptoms and recurrence, surgical intervention is often complicated by tumor size, and consequently, complex distortion of normal anatomy [4].

In this study, we aim to detail and provide an update on our institution’s experience with the surgical management of both sacral and presacral schwannomas [6]. A comprehensive literature review was conducted to characterize surgical sacral schwannomas and their postoperative outcomes.

Methods

Patient cases

We retrospectively reviewed the records of patients who underwent surgery for the removal of sacral or presacral schwannoma at our institution—Mayo Clinic (Minnesota, Arizona, and Florida)—between 1997 and 2018 following Institutional Review Board (IRB) approval (IRB number 21-000125). We abstracted the following information: demographics, presenting symptoms, lesion characteristics, surgical management, and outcomes. Patient demographic data included age and gender. Lesion characteristics included size, need for lumbosacral laminectomy, nerves involved and laterality. Surgical outcomes included progression, improvement, follow-up, and surgical complications. Surgical management details included operative approach taken to remove the lesion (anterior, posterior, or combined), nerve sacrifice, how extensive the surgical resection was, and whether a revision surgery was necessary. Patient outcomes were characterized by lesion progression (determined by radiology reports), range of recovery (full, partial or none), follow-up time, survival, and postoperative complications.

Study selection

We conducted a systematic review of peer-reviewed articles from inception to December 2020 using relevant word searches performed on PubMed, EMBASE, Web of Science, Scopus, Medline, and Cochrane Library. To maximize search results, specific keywords were used as Medical Subject Headings (MeSH) terms in all logical permutations to identify relevant studies: “spinal OR spine,” “schwannoma,” “sacral,” “presacral” and “surgical treatment.” The results were screened by two authors to determine eligibility for inclusion in final review. Eligibility criteria included original research in English involving human subjects. Narrative reviews, abstracts, book chapters, and cadaveric studies were excluded. Studies involving non-sacral schwannomas or patients not treated surgically, and studies that were not full-length articles, were also excluded.

Data collection and statistical analysis

Data were extracted from 31 relevant studies and tabulated regarding patient age and gender, presenting symptoms, size and location of tumor, and surgical management and outcome including complications and/or recurrence. Analysis through an unpaired two-sample t-test was used to observe associations of continuous variables between different groups. Fisher’s exact test was utilized to assess statistical significance between categorical variables. Kaplan–Meier curve demonstrating disease-free survival was generated in R4.1.2 using the survival package. All statistical analyses were conducted using the R language (version 4.1.2) and RStudio. P values less than 0.05 were considered significant.

Results

Demographics

We identified 27 patients with sacral schwannomas at our institution. Average age was 49.9 years, and 44.4% identified as female (Table 1). In the literature, 93 patients were identified with an average age of 43.4 years and 60.6% identifying as female [5, 7–36]. None of our patients were known to have schwannomatosis or neurofibromatosis type 2. Only one patient in the literature, the patient in the case report by Braley et al., had a history of schwannomatosis while no patients had any known history of neurofibromatosis type 2.

Presenting symptoms

Most patients at our institution (88.9%) and in the literature (78.7%) experienced pain related to their schwannomas (Tables 1 and 2). Sexual dysfunction was the least common symptom experienced by our patients and those in the literature (3.7% and 3.2%, respectively). Other symptoms including weakness, sensory changes, and bowel/bladder incontinence had more variable distributions amongst both patient populations. Intraoperative monitoring was included for 15 (55.6%) of our patients (including anal sphincter monitoring for six patients). At our institution, 29.6% of patients experienced weakness, 33.3% sensory changes, and 37% bowel/bladder incontinence. In the literature, 12.8% of patients experienced weakness, 20.2% sensory changes and 26.6% bowel/bladder incontinence.
Lesion characteristics

The dimensions for the largest lesion surgically treated at our institution were $11.5 \times 7 \times 3.5$ cm (Fig. 1). The largest lesion found in the literature was considerably greater, measuring $12.1 \times 11 \times 10.7$ cm. Laminectomies were performed more often for patients treated at our institution (44.4% at S1-S2 level and 33.3% at L5-S1) than for patients in the literature (10.6% at S1-S2 level and 9.6% at L5-S1). Involvement of the S1 nerve was the most frequent for our patients (70.3%). However, only 26.6% of cases in the literature demonstrated S1 nerve involvement. While less common, the L5 nerve showed a similar trend, with involvement in 22.2% of our cases but only 8.5% of cases in the literature. Tumor predilection for right, left or combined laterality was well-distributed. At our institution, 48.1% of tumors were right-sided, 37% left-sided, and 11.1% bilateral. In the literature, 14.9% of tumors were right-sided, 17.0% left-sided, and 10.6% bilateral. In our cohort, patients with sacral tumors without extension into the presacral space outnumbered those extending into it (15 versus 8) while there was an equal number of patients with presacral tumors either limited to or extending into the foramina or sacrum (two patients in both). Six of our patients had schwannomas that were intradural.

Surgical management

Most surgeries performed on our patients (59.3%) and on those in the literature (51.1%) were through a posterior approach. An anterior approach was used for 22.2% of our patients and 28.7% of patients in the literature (Table 3). The least common technique used in both patient populations was a combined anterior and posterior surgery (14.8% of our patients and 18.1% of patients in the literature). Most patients had gross total resection (63% of ours and 72.3% in the literature). Subtotal resections were performed on 40.7% of our patients and 26.6% of those in the literature. Furthermore, 33.3% of patients treated at our institution had a nerve sacrificed and 40.7% required a revision surgery. In the literature, however, only 4.3% of patients treated had a nerve sacrificed, and 8.5% underwent revision surgery.

Outcomes

The average follow-up time for our patients was 43 months, compared to 60.1 months for patients in the literature (Table 3). Moreover, 59.3% of our patients reported full recovery from preoperative symptoms while only 31.9% of those in the literature recovered fully. However, fewer patients in the literature reported no recovery of symptoms (1.1%) compared to our patient cohort (11.1%). An approximately even number of patients in both populations reported a partial recovery (33.3% at our institution and 29.8% in the literature).

More patients experienced lesion progression at our institution (25.9% versus 14%) and had a shorter time to progression (37.8 months versus 60.2 months). Furthermore, our patients demonstrated a lower complication rate
Table 2 Baseline Characteristics, clinical diagnosis/symptoms, and pathological features of tumors in patients found in the literature who underwent sacral schwannoma resection

| Study and year                  | Total (N) | Baseline characteristics | Presenting symptoms | Lesion characteristics | Nerve(s) involved | Laterality |
|--------------------------------|-----------|--------------------------|---------------------|------------------------|------------------|------------|
|                                |           | Age                      | Female Sex          | Pain                    | Weakness         | Sensory changes | Bowel/Bladder incontinence | Sexual dysfunction | Nerve(s) involved | Laterality |
| Abernathy et al., 1986         | 13        | 38.6 ± 12.8 [16–57]      | 6 (46.2%)           | 12 (92.3%)             | 2 (15.4%)        | 1 (7.7%)     | 2 (15.4%)                  |                  | L5 involved in 2 patients (15.4%) | –               |
|                                |           |                          |                     |                        |                  |             |                          |                  | S1 involved in 1 patient (7.7%) | –               |
| Accicarri et al., 1996         | 1         | 19                       | Yes                 | –                      | –                | –            | –                        |                  | S2               | Right       |
| Attiah et al., 2015            | 1         | 58                       | Yes                 | Yes                    | Yes             | –            | –                        |                  | S2               | Bilateral   |
| Braley et al., 2020            | 1         | 67                       | Yes                 | Yes                    | Yes             | Yes          | –                        |                  | S1               | Left        |
| Cagli et al., 2012             | 13        | 37.2 ± 13.6 [14–55]      | 8 (61.5%)           | 13 (100%)              | 3 (23.1%)       | –            | 4 (30.8%)                  | 1 (7.7%)          | 1 S2             | Bilateral   |
| Camacho et al., 2019           | 1         | 58                       | Yes                 | Yes                    | –                | –            | –                        |                  | S1               | –           |
| Chandhanayingyong et al., 2008 | 4         | 45.5 ± 14.5 [29–62]      | 3 (75%)             | 4 (100%)               | –                | 2 (50%)     | 1 (25%)                  |                  | S1 involved in 3 patients (75%) | 1 Left (25%)   |
|                                |           |                          |                     |                        |                  |             |                          |                  | S2 involved in 4 patients (100%) | 1 Right (25%) |
|                                |           |                          |                     |                        |                  |             |                          |                  | S3 involved in 1 patient (25%) | 1 Bilateral (25%) |
|                                |           |                          |                     |                        |                  |             |                          |                  | S4 involved in 1 patient (25%) | –               |
| Dominguez et al., 1997         | 6         | 40.7 ± 19.8 [17–68]      | 5 (83.3%)           | 4 (66.7%)              | –                | 2 (33.3%)   | 4 (66.7%)                  | –                | –               | –           |
| Emohare et al., 2015           | 1         | 49                       | 0                   | Yes                    | –                | –            | Yes                      | No               | S1-S2            | Left        |
| Gethardt et al., 2020          | 1         | 49                       | 0                   | Yes                    | –                | –            | –                        |                  | S2-S4            | Right       |
| Higgin et al., 2014            | 1         | 71                       | 0                   | –                      | –                | –            | –                        |                  | L4-S1            | Left        |
| Huang et al., 2020             | 1         | 34                       | 0                   | –                      | –                | –            | –                        |                  | –               | –           |
| Kanamori et al., 2013          | 1         | 58                       | Yes                 | Yes                    | Yes             | –            | –                        |                  | S1-S3            | Left        |
| Khan et al., 2018              | 1         | 38                       | 0                   | Yes                    | –                | Yes          | –                        |                  | S1-S2            | Bilateral   |
| Lecerc et al., 2020            | 6         | 52 ± 15.6 [32–69]        | 3 (50%)             | 3 (50%)                | –                | 1 (16.7%)   | 3 (50%)                  | –                | S1 involved in 3 patients (50%) | 2 Left (33.3%) |
|                                |           |                          |                     |                        |                  |             |                          |                  | S2 involved in 2 patients (33.3%) | 4 Right (66.7%) |
|                                |           |                          |                     |                        |                  |             |                          |                  | S3 involved in 1 patient (16.7%) | –               |
| Lee_1 et al., 2017             | 1         | 40                       | Yes                 | Yes                    | Yes             | –            | –                        |                  | S2               | Left        |
| Lee_2 et al., 2017             | 1         | 47                       | Yes                 | Yes                    | Yes             | –            | –                        |                  | S1               | Left        |
| Lin et al., 2016               | 1         | 23                       | Yes                 | Yes                    | –                | Yes          | –                        |                  | S1-S3            | Bilateral   |
| Maccio et al., 2019            | 1         | 62                       | Yes                 | Yes                    | –                | –            | –                        |                  | –               | Right       |
| Masanobu et al., 2001          | 1         | 45                       | No                  | Yes                    | Yes             | –            | –                        |                  | S1-S3            | Bilateral   |
| Study and year          | Total (N) | Age       | Female Sex | Presenting symptoms | Lesion characteristics |
|------------------------|-----------|-----------|------------|----------------------|------------------------|
|                        |           |           |            |                      | Nerve(s) involved      | Laterality             |
|                        |           |           |            |                      |                        |                        |
| Mohanty et al., 2018   | 9         | 43 ± 11.8 [19–63] | 6 (66.7%) | 9 (100%) 1 (11.1%) – | 1 (11.1%) – S1 involved in 2 patients (22.2%) | –                       |
|                        |           |           |            |                      | S2 involved in 5 patients (55.6%) |                        |
|                        |           |           |            |                      | S3 involved in 8 patients (88.9%) |                        |
|                        |           |           |            |                      | S4 involved in 8 patients (88.9%) |                        |
|                        |           |           |            |                      | S5 involved in 4 patients (44.4%) |                        |
| Ortolan et al., 1996   | 1         | 27        | Yes        | Yes – Yes – – –      | L5-S1 Right            |                        |
| Oshima et al., 2004    | 1         | 54        | Yes        | Yes Yes Yes Yes –    | S1 Right               |                        |
| Pennington et al., 2019| 7         | 39.4 ± 24.5 [10–73] | 3 (42.9%) | 4 (57.1%) – – 2 (28.6%) 1 (14.3%) | S1 involved in 1 patient (14.3%) | 2 Right (28.6%)        |
| Pongsthorn et al., 2009| 6         | 49.3 ± 7.8 [38–58] | 4 (66.7%) | 6 (100%) 4 (66.7%) 1 (16.7%) – | S1 involved in 2 patients (33.3%) | 2 Right (33.3%)        |
|                        |           |           |            |                      | L5 involved in 1 patient (16.7%) |                        |
| Ragurajaprakash et al., 2020 | 1         | 56        | Yes        | Yes Yes Yes Yes –    | – Bilateral            |                        |
| Silva et al., 2018     | 1         | 1         | No         | Yes – – – – –        | L5-S1 Left             |                        |
| Tahta et al., 2020     | 1         | 46        | No         | – – – Yes –          | S1 Left                |                        |
| Torgal et al., 2014    | 1         | 42        | No         | Yes Yes – –          | L5-S1 Left             |                        |
| Yang et al., 2007      | 1         | 67        | No         | Yes – – Yes –        | Sciatic Left           |                        |
| Yin et al., 2018       | 7         | 45 ± 15 [25–65] | 6 (85.7%) | – – – – – – – – – – | – – – – – – – – – – | – – – – – – – – – – |
(14.8%) compared with patients in the literature (25.5%). Complications at our institution were neurological in origin and included neuropathic pain, dysesthesia, paraplegia, and incontinence. The survival rate was 92.6% for our patients and 93.6% for patients in the literature. Patient-level data may provide insight into each patient’s course (see Supplemental Tables 1–4).

**Statistical analysis**

Kaplan–Meier curves for disease-free survival for all patients showed no significant variation in progression by extent of surgical resection (log-rank \( P = 0.26 \); see Supplemental Fig. 1). No significant variation in progression of spinal schwannoma between gross total and subtotal resection was found in analysis of only patients found in the literature (see Supplemental Fig. 2). Univariate analysis demonstrated that patients with no lesion progression were more likely to have full or partial improvement in symptoms (\( p = 0.044 \)) and that female patients were more likely to undergo resection via a posterior approach (\( p = 0.042 \)) (Table 4; significant values are in bold). No other relationships were found to be statistically significant.

**Discussion**

**Epidemiology and clinical presentation**

The fourth and fifth decades of life are the typical age range when schwannomas are diagnosed, consistent with patients at our institution and those in our literature search. The literature also indicates that Black and American Indian/Alaska Native races were associated with lower incidence of spinal schwannomas when compared to Whites and Asians [37]. While schwannomas may occur in the sacral region of the spinal canal, they comprise a small percentage of the variety of neoplasms that may arise in this location; other tumors found to occur in this region include chordomas, chondrosarcomas, neurofibromas, malignant peripheral nerve sheath tumors (MPNSTs), giant cell tumors, plasmacytomas, lymphomas, aneurysmal bone cysts, inflammatory and congenital lesions. The most consistent presenting symptom (88.9% of our patients versus 78.7% in the literature) was local pain. Sexual dysfunction was relatively uncommon, with only 3.7% and 3.2% of patients experiencing it both at our institution and in the literature, respectively.

Accurately diagnosing the cause of low back pain and the appropriate treatment thereafter is important in improving patient prognosis and reducing costs, as low back pain affects most people at least once in their lifetime. Kim et al. reported a case in which a sacral schwannoma was discovered with MRI of the sacrolumbar region following a lumbar epidural block performed for low back pain of a few years’ duration. Imaging was ordered only when the patient presented with transient cauda equina syndrome—perineal numbness, lower extremity weakness, and decreased deep-tendon reflexes—that completely resolved over the following 9 h before discharge home [38]. This was triggered by the lumbar epidural

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**Fig. 1**  
A Preoperative T1 MRI for patient 1 at our institution showing a large (11.5 × 7 × 3.5 cm) right-sided sacral schwannoma invading the abdominal cavity.  
B Postoperative T1 MRI following the removal of the tumor via transsacral approach
Table 3  Surgical characteristics and postoperative outcomes of patients found in the literature who underwent sacral schwannoma resection

| Study and year       | Total (N) | Surgical management | Postoperative outcomes and follow-up |
|----------------------|-----------|---------------------|--------------------------------------|
|                      |           | Operative approach  | Sacrifice of nerve(s) | Extent of surgical resection | Revision surgery | Lesion progression | Time to progression (months) [range] | Improvement of symptoms | Follow-up time (months) | Survival | Complications |
| Abernathy et al., 1986 | 13        | 3 Anterior (23.1%)  | –                      | 4 Gross total (30.8%)             | 4 (30.8%)         | 2 (15.4%)          | 94.5 ± 103.9 [21–168]               | 8 Full recovery (61.5%) | 109.1 ± 120.3 [5–399]     | Yes       | 11 Yes        |
|                      |           | 10 Posterior (76.9%)|                       | 9 Subtotal (69.2%)               |                  |                   |                                  |                          |                        | 2 No       |
|                      |           |                     |                        |                                   |                   |                   |                                  |                          |                        | (unrelated cause)        |
| Accicarri et al., 1996 | 1         | Anterior            | –                      | Gross total                       | No                | No                | N/A                              | Partial recovery         | 6           | Yes         | No          |
| Attiah et al., 2015  | 1         | Posterior           | Yes                    | Gross total                       | No                | No                | N/A                              | Partial recovery         | 1           | Yes         | No          |
| Braley et al., 2020  | 1         | Posterior           | –                      | Gross total                       | No                | No                | N/A                              | Partial recovery         | –           | Yes         | No          |
| Cagli et al., 2012   | 13        | 2 Anterior (15.4%)  | –                      | Gross total                       | 1 (7.7%)          | 2 (15.4%)          | 78 ±42.4 [48–108]                | Partial recovery         | 105.2 ± 56.5 [24–204]     | Yes       | 5 (38.5%)    |
|                      |           | 6 Posterior (46.2%) |                        |                                   |                   |                   |                                  |                          |                        |                        |
|                      |           | 5 Combined (38.5%)  |                        |                                   |                   |                   |                                  |                          |                        |                        |
| Camacho et al., 2019 | 1         | Posterior           | –                      | Gross total                       | No                | No                | N/A                              | Partial recovery         | 12          | Yes         | No          |
| Chandhanayaying et al., 2008 | 4       | Posterior           | –                      | 2 Gross total (50%)               | No                | 1 (25%)           | 14 ±9.9 [7–21]                  | 3 Full recovery (75%)     | 18.3 ± 8.4 [7–27]         | Yes       | 1 (25%)      |
| Dominguez et al., 1997 | 6        | 4 Posterior (66.7%) | –                      | Gross total                       | 1 (16.7%)         | 1 (16.7%)         | 204                              | 5 Full recovery (83.3%)     | 9.2 [18–252]            | Yes       | –           |
|                      |           | 2 Combined (12.5%)  |                        |                                   |                   |                   |                                  |                          |                        |                        |
| Emohare et al., 2015 | 1         | Lateral             | No                     | Gross total                       | No                | No                | N/A                              | Full recovery             | 1           | Yes         | No          |
| Gerhardt et al., 2020 | 1         | Anterior            | No                     | Gross total                       | No                | No                | N/A                              | Full recovery             | 29          | Yes         | No          |
| Higgin et al., 2014  | 1         | Anterior            | –                      | Gross total                       | No                | No                | N/A                              | –                           | 3           | Yes         | No          |
| Huang et al., 2020   | 1         | Anterior            | –                      | Gross total                       | No                | No                | N/A                              | –                           | 18          | Yes         | No          |
| Kanamori et al., 2013 | 1        | Posterior           | No                     | Subtotal                          | No                | No                | N/A                              | –                           | 36          | Yes         | –           |
| Study and year | Total (N) | Surgical management | Postoperative outcomes and follow-up |
|---------------|-----------|---------------------|--------------------------------------|
|               |           | Operative approach  | Sacrifice of nerve(s) | Extent of surgical resection | Revision surgery | Lesion progression | Time to progression (months) [range] | Improvement of symptoms | Follow-up time (months) | Survival | Complications |
| Khan et al., 2018 | 1         | Combined            | No                      | Gross total                     | No              | No              | N/A                       | Full recovery           | –              | Yes       | No          |
| Leclerc et al., 2020 | 6         | Anterior            | No                      | 1 Gross total (16.7%)             | Yes             | Yes             | 67 ± 6.39 [7–165]         | Full recovery           | 5               | 1 (16.7%) |
| Lee_1 et al., 2017 | 1         | Posterior           | No                      | Gross total                     | No              | No              | N/A                       | Full recovery           | 24             | Yes       | No          |
| Lee_2 et al., 2017 | 1         | Anterior            | –                       | Gross total                     | No              | –               | N/A                       | Partial recovery        | 24             | Yes       | –           |
| Lin et al., 2016  | 1         | Posterior           | No                      | Gross total                     | No              | No              | N/A                       | Partial recovery        | 12             | Yes       | No          |
| Maccio et al., 2019 | 1         | Anterior            | No                      | Gross total                     | No              | No              | N/A                       | Full recovery           | 18             | Yes       | Yes         |
| Masanobu et al., 2001 | 1         | Combined            | No                      | Gross total                     | No              | No              | N/A                       | Partial recovery        | 24             | Yes       | No          |
| Mohanty et al., 2018 | 9         | Posterior           | –                       | 6 Gross total (66.7%)             | 4 (44.4%)       | 23 ± 25.5 [2–60] | 44.8 ± 29.3 [9–96]       | 7 (77.8%)               | No             |           |
| Ortolan et al., 1996 | 1         | Posterior           | Yes                     | Gross total                     | No              | No              | N/A                       | Partial recovery        | 17             | Yes       | No          |
| Oshima et al., 2004 | 1         | Combined            | Yes                     | Gross total                     | No              | No              | N/A                       | Partial recovery        | 3              | Yes       | –           |
| Pennington et al., 2019 | 7         | 3 Anterior (42.9%)  | No                      | 6 Gross total (85.7%)             | 1 (14.3%)       | 30               | –                        | 14.2 ± 13.2 [0–83]      | 6              |           |
| Pongthorn et al., 2009 | 6         | 1 Anterior (16.7%)  | 1 (16.7%)               | 3 Gross total (50%)              | 1 (16.7%)       | 84               | –                        | 86 ± 74.6 [6–180]       | Yes            | 2 (33.3%) |
| Ragurajaparakash et al., 2020 | 1         | Anterior            | –                       | Subtotal                        | No              | No              | N/A                       | Full recovery           | –              | Yes       | –           |
block meant to relieve the refractory back and leg pain the patient had been experiencing for years.

Treatment algorithm

Various surgical approaches in treating spinal schwannomas have been developed. Gross total resection was seen more often in the literature (72.3%) compared to the retrospective data at our institution (63%) and likely explains the difference in need for revision surgery (40.7% versus 8.51% in the literature) and subsequent tumor progression (25.9% versus 14% in the literature). However, most patients at our institution fully recovered (59.3%) as opposed to those in the literature (31.9%). Still, overall survival was high for both patient populations (92.6% versus 93.6% in the literature). While total resection is often pursued in the literature, it is not always successful given the variable tumor sizes. The risk of developing neurologic deficits from total resection exists alongside the benefit of preventing recurrence [39–41].

During surgical removal of schwannoma, care must be taken to avoid damaging the surrounding neurovascular organs. Importantly, of the nine patients at our institution presenting with bowel/bladder incontinence, five (55.6%) were diagnosed with schwannomas involving the S2 nerve, thus increasing the risk of dysfunction of the pudendal nerve. Four of the nine patients presenting with incontinence had schwannomas involving the S1 nerve but none of the lower sacral nerves; three of them presented with incontinence due to mechanical displacement i.e., tumor putting pressure on either the bladder or colon, while one had longstanding incontinence due to a history of transverse myelitis. None of our patients suffered from bowel or bladder incontinence as a postoperative complication. The most frequent operative approach employed was from the posterior (59.3% versus 51.1% in the literature), which is reasonable as it avoids the need to dissect through the abdomen and is optimal for cases in which the tumor extends into the spinal canal or sacrum with a small presacral component. However, the treatment plan of surgical approaches varies depending on the intrasacral and retroperitoneal extension of the mass [42]. An anterior transabdominal or retroperitoneal approach may be employed to protect the vascular plexus and intrapelvic organs while liberating the tumor. When the tumor is limited to the front of the sacrum, an anterior approach may be preferred. Otherwise, these tumors may be removed via the posterior approach alone. Moreover, a posterior approach with proper fenestration may be used to remove sacral schwannomas with large presacral components. A combined approach may be worth considering to allow for complete resection when a schwannoma consists of an extraspinal portion larger than the intraspinal and vertebral body portions or erosion of the lumbar vertebral body [43].

Table 3 (continued)

| Study and year | Operative management | Surgical approach | Sacrifice of nerve(s) | Extent of surgical resection | Lesion progression | Time to progression (months) | Improvement of Symptoms | Survival | Complications |
|---------------|---------------------|------------------|----------------------|-----------------------------|-------------------|-----------------------------|------------------------|----------|---------------|
| Silva et al., 2018 | 1 | Posterior | No | Gross total | No | N/A | N/A | No | N/A | – | 24 | Yes | – |
| Tahta et al., 2020 | 1 | Combined | No | Gross total | No | N/A | N/A | No | N/A | N/A | Full recovery | 12 | Yes | No |
| Torgal et al., 2014 | 1 | Combined | No | Gross total | No | N/A | N/A | No | N/A | N/A | Full recovery | 12 | Yes | Yes |
| Yang et al., 2007 | 1 | Anterior | No | Gross total | No | N/A | N/A | No | N/A | N/A | Full recovery | 6 | Yes | No |
| Yin et al., 2018 | 7 | Anterior (71.4%) | No | Gross Total | No | N/A | N/A | No | N/A | N/A | Full recovery | 24–31 | Yes | No |
Table 4 Univariate analysis of patient demographics, symptomology, surgery, and clinical course stratified by extent of resection, lesion progression, and surgical approach

|                          | Extent of resection | Lesion progression | Surgical approach |
|--------------------------|---------------------|--------------------|-------------------|
|                          | Gross total (N=16)  | Subtotal (N=11)    | Total (N=27)      | P Value     | No lesion progression (N=20) | Lesion progression (N=7) | Total (N=27) | P value | Anterior (N=6) | Combined (N=4) | Posterior (N=15) | Total (N=25) | P value |
| Mean age (SD)            |                     |                    |                   |             |                       |                         |                |         |                |                 |                |               |         |        |
|                          | 48.000 (9.832)      | 52.545 (13.699)    | 49.852 (11.538)   | 0.324       | 48.850 (10.835)       | 52.714 (13.865)          | 49.852 (11.538) | 0.46    | 49.000 (7.797) | 52.000 (12.910) | 48.133 (12.478) | 48.960 (11.238) | 0.84    |
| Female sex               |                     |                    |                   |             |                       |                         |                |         |                |                 |                |               |         |        |
|                          | 8 (50.0%)           | 4 (36.4%)          | 12 (44.4%)        | 0.696       | 8 (40.0%)             | 4 (57.1%)               | 12 (44.4%)     | 0.66    | 5 (83.3%)      | 0 (0.0%)        | 7 (46.7%)       | 12 (48.0%) | 0.042  |
| Pain                     |                     |                    |                   |             |                       |                         |                |         |                |                 |                |               |         |        |
|                          | 14 (87.5%)          | 9 (81.8%)          | 23 (85.2%)        |             | 17 (85.0%)            | 6 (85.7%)              | 23 (85.2%)     | 1       | 6 (100.0%)     | 4 (100.0%)      | 11 (73.3%)      | 21 (84.0%) | 0.32   |
| Weakness                 |                     |                    |                   |             |                       |                         |                |         |                |                 |                |               |         |        |
|                          | 5 (31.2%)           | 3 (27.3%)          | 8 (29.6%)         | 1           | 4 (20.0%)             | 4 (57.1%)              | 8 (29.6%)      | 0.15    | 2 (33.3%)      | 0 (0.0%)        | 5 (33.3%)       | 7 (28.0%) | 0.58   |
| Sensory deficit          |                     |                    |                   |             |                       |                         |                |         |                |                 |                |               |         |        |
| Bowel/Bladder incontinence |                 |                    |                   |             |                       |                         |                |         |                |                 |                |               |         |        |
| Sexual dysfunction       |                     |                    |                   |             |                       |                         |                |         |                |                 |                |               |         |        |
| Improvement of symptoms  |                     |                    |                   |             |                       |                         |                | 0.044   | 0 (0.0%)       | 0 (0.0%)        | 1 (6.7%)       | 1 (4.0%) | 0.1    |
| No recovery              | 2 (12.5%)           | 1 (9.1%)           | 3 (11.1%)         |             | 1 (5.0%)              | 2 (28.6%)              | 3 (11.1%)      | 0 (0.0%) | 2 (50.0%)     | 1 (6.7%)        | 1 (12.0%)       | 3 (12.0%) | 0.054  |
| Full recovery            | 9 (56.2%)           | 6 (54.5%)          | 15 (55.6%)        |             | 10 (50.0%)            | 5 (71.4%)              | 15 (55.6%)     | 2 (33.3%) | 2 (50.0%)     | 9 (60.0%)       | 13 (52.0%)      | 0.65    |
| Partial recovery         | 5 (31.2%)           | 4 (36.4%)          | 9 (33.3%)         |             | 9 (45.0%)             | 0 (0.0%)               | 9 (33.3%)      | 4 (66.7%) | 0 (0.0%)      | 5 (33.3%)       | 9 (36.0%)       | 0.054   |
| L5 nerve involvement     | 5 (31.2%)           | 1 (10.0%)          | 6 (23.1%)         | 0.352       | 4 (21.1%)             | 2 (28.6%)              | 6 (23.1%)      | 1       | 3 (50.0%)      | 1 (25.0%)       | 1 (7.1%)        | 5 (20.8%) | 0.054  |
| S1 nerve involvement     | 13 (81.2%)          | 7 (70.0%)          | 20 (76.9%)        | 0.644       | 14 (73.7%)            | 6 (85.7%)              | 20 (76.9%)     | 1       | 5 (83.3%)      | 3 (75.0%)       | 10 (71.4%)      | 18 (75.0%) | 1      |
| S2 nerve involvement     | 8 (50.0%)           | 6 (60.0%)          | 14 (53.8%)        | 0.701       | 10 (52.6%)            | 4 (57.1%)              | 14 (53.8%)     | 1       | 3 (50.0%)      | 1 (25.0%)       | 8 (57.1%)       | 12 (50.0%) | 0.65   |
| Number of nerve levels involved | 0.731       | 0.84               | 0.91             | 0.284       | 1                     | N/A                    |
Table 4 (continued)

| Extent of resection | Gross total (N = 16) | Subtotal (N = 11) | Total (N = 27) | P Value | Lesion progression | Total (N = 27) | P Value | Surgical approach | Anterior (N = 6) | Combined (N = 4) | Posterior (N = 15) | Total (N = 25) | P value |
|---------------------|----------------------|------------------|----------------|---------|-------------------|----------------|---------|-------------------|-----------------|-----------------|-------------------|----------------|---------|
| Anterior            |                      |                  |                |         |                   |                |         |                   |                 |                 |                   |                |         |
| Combined            |                      |                  |                |         |                   |                |         |                   |                 |                 |                   |                |         |
| Posterior           |                      |                  |                |         |                   |                |         |                   |                 |                 |                   |                |         |
| Extent of resection |                      |                  |                |         |                   |                |         |                   |                 |                 |                   |                |         |
| Gross total         | N/A                  | N/A              | N/A            |         |                   |                |         |                   |                 |                 |                   |                | 0.28    |
| Subtotal            | N/A                  | N/A              | N/A            |         |                   |                |         |                   |                 |                 |                   |                |         |
| Complications       | 1 (6.2%)             | 3 (27.3%)        | 4 (14.8%)      | 0.273   |                   |                |         |                   |                 |                 |                   |                |         |
| Lesion Progression  | 4 (25.0%)            | 3 (27.3%)        | 7 (25.9%)      | 1       | N/A               | N/A            |         |                   |                 |                 |                   |                |         |
| Mean time to        | 33.425 (22.266)      | 43.567 (40.035)  | 37.771 (28.488)| 0.683   |                   |                |         |                   |                 |                 |                   |                |         |
| progression in      |                      |                  |                |         |                   |                |         |                   |                 |                 |                   |                |         |
| months (SD)         |                      |                  |                |         |                   |                |         |                   |                 |                 |                   |                |         |
| Mean follow-up      | 19.875 (32.660)      | 62.227 (85.273)  | 37.130 (62.144)| 0.082   |                   |                |         |                   |                 |                 |                   |                |         |
| time in months      |                      |                  |                |         |                   |                |         |                   |                 |                 |                   |                |         |
| (SD)                |                      |                  |                |         |                   |                |         |                   |                 |                 |                   |                |         |
| Revision surgery    | 4 (66.7%)            | 1 (25%)          | 5 (33.3%)      | 0.44    |                   |                |         |                   |                 |                 |                   |                |         |

The bolded values are p values <0.05, indicating significance
Because local recurrence and malignant transformation are very rare, subtotal resection or simple enucleation is frequently the preferred treatment of choice. The risk that it can regrow still exists though, and if removed inadequately, the reoperations have higher complication risks. A postoperative CT scan may aid in planning the reconstruction of bone structures depending on the destruction of the sacral bone and the invasion of the sacroiliac joint.

**Patient outcomes**

Patients with sacral or presacral schwannomas at our institution had a shorter average follow-up time and a lower complication rate than those in the literature. Also, more of our patients reported full recovery from preoperative symptoms. However, more of our patients reported no recovery of symptoms. Notably, more patients at our institution experienced lesion progression, which may be due to a higher rate of postoperative MRI with our patients relative to those treated at other institutions.

Clinical outcomes of patient studies following sacral schwannoma resection have been characterized for significance. One study by Pan et al. assessed ten patients between the ages of 31 and 63 years old. One patient underwent an anterior approach, eight patients followed a posterior approach, and two patients underwent a combined approach. The results found the average surgical blood loss at 980 mL, with three patients suffering from postoperative complications such as bladder/bowel dysfunction and CSF leakage with secondary intracranial infection. Six patients underwent biopsies with no subsequent complications afterwards, with the overall average follow-up time being 22.7 months [43].

Another study by Pongstorm et al. treated six cases of giant sacral schwannoma. The average patient age was 47.8 years, with all patients having the same clinical presentation of lumbosacral pain. The surgical approach was posterior in two patients, anterior in one, and combination in three. The mean surgical time was 7.8 h with an average blood loss of 2562 g. Only one patient required a second surgery. One patient had postoperative complications of erectile dysfunction and motor weakness, while another had causalgia in the right leg. During final follow-up, no patients presented with pain or neurological deficits due to surgical treatment. Piecemeal subtotal excision was found to have positive outcomes [31].

A study by Chandhanayingyon et al. further assessed sacral schwannoma removal using intralesional curettage and adjuvant radiation therapy. The study involved four cases, three females and one male, with an average age of 45.5 years. The primary symptom was lumbosacral pain, with each patient undergoing surgery through a posterior approach. Final follow-up found lumbosacral relief in all patients and no neurological deficits or recurrent symptoms.

Radiographic imaging found marginal sclerosis at the lesion site for one patient. However, it was still found that intrasional curettage and adjuvant radiation therapy effectively relieved sacral schwannoma symptoms [13].

Sowash et al. conducted a retrospective review of thirty-two patients with giant sacral schwannomas. Sixteen cases used the posterior approach and three underwent the combined approach, with instrumentation placed in ten cases. Gross tumor resection was achieved in 19 patients, with 12 showing enhancement on MRI following surgery. Five patients experienced complications, including chemical meningitis, wound infections, gastrointestinal obstruction, and Guillain-Barré Syndrome. Long-term follow-up showed all patients improved with regards to nonradicular pain, sensory deficits, and bowel and bladder function. Three patients showed tumor recurrence, yet surgical resection of sacral schwannoma was overall found to improve clinical outcomes [44]. In a case series by Handa et al., eleven patients with giant schwannomas were treated surgically. Four were treated posteriorly, three anteriorly, and four through a combined approach. Average blood loss was 3740 g, and three patients experienced complications. Complications included massive bleeding, causalgia, and motor weakness. Recurrence occurred in two patients, and one patient required another surgery [45]. These studies all indicate positive clinical outcomes in improving symptom presentation without manifesting neurological deficits or complications. The posterior approach was the most widely used approach, with blood loss varying between studies. While the occurrence of complications and tumor recurrence is possible, most surgical treatments for sacral schwannoma are effective.

**Limitations**

This analysis includes studies with low-level evidence and no prospective or randomized control trials, limiting the strength of conclusions from our qualitative and quantitative analysis. We were not able to perform a survival analysis as this type of tumor is benign, and most of the cases were resolved surgically. Furthermore, we could not assess the impact of radiotherapy on survival. Another limitation is that our literature search was confined to major databases and English studies. Conclusions drawn from our Kaplan–Meier and univariate analyses require further investigation given the rarity of this disease and the limited amount of data available. Finally, we could not study the cause-and-effect relationships and assess the rate of this disease because there was no relevant comparative group.
Conclusion

Sacral schwannomas are uncommon benign tumors of the spine, which may require operative management. Patient experience can vary significantly in terms of demographics, symptoms, tumor qualities, surgical treatment protocol, and outcome. Among the range of symptoms experienced, the most common was pain. Most patients were treated through gross total resection via the posterior approach. The prognosis may be good, with preservation of neurologic function and a high overall survival rate, when the surgical approach is well-planned and well-executed.

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Declarations

Conflict of interest The authors have no relevant financial or non-financial interests to disclose.

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