Original Article

DOI: 10.7860/JCDR/2026/78962.22731

Clinical Profile and Surgical Outcomes of Giant Presacral Schwannomas: A Retrospective Descriptive Study from Tamil Nadu, India

Surgery Section

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ABSTRACT

Introduction: Presacral tumours are an extremely rare entity. Schwannomas account for only 3% of tumours in this region. Schwannomas are benign, unifocal and often asymptomatic. Presacral lesions are usually asymptomatic, although a few of these patients may present with compressive symptoms or neurogenic pain. Diagnosis and characterisation of nerve sheath tumours, of which schwannoma is a part, is usually done with Magnetic Resonance Imaging (MRI). Surgical excision is the mainstay of management, although it is associated with high morbidity. Although a few case reports are available, large datasets are sparse.

Aim: To study the clinical presentation, imaging features and management of patients who underwent excision of presacral schwannomas.

Materials and Methods: The present retrospective descriptive study was conducted in General Surgery Unit-IV in Christian Medical College and Hospital, Vellore, Tamil Nadu, India, from July 2013 to July 2024. All patients above 18 years of age, histopathologically confirmed presacral schwannomas, were identified and included. The demographic profile, presenting symptoms, imaging findings, preoperative assessment, surgical details, postoperative complications, and follow-up were recorded. Descriptive statistics were calculated, with mean and median serving as the measure of central tendency and the standard deviation representing the measure of dispersion.

Results: A total of 20 patients were identified with a mean age of 41.3 years (±14.05), with 13 patients being male and the remaining female. Eleven patients presented with pressure symptoms due to direct compression of the pelvic organs. Imaging of choice was MRI in 19 patients. Seventeen patients had preoperative biopsy done, of which 16 were schwannoma and one was neurofibroma. Median largest diameter of these tumours at the time of presentation was 10.55 cm with a range of 6.8 to 25 cm. 19 patients underwent a laparotomy (transperitoneal approach) with intracapsular excision of the tumour. The mean operating time was 2.85 hours (±1.12), with a mean blood loss of approximately 500 mL (±240). 10% of patients developed postoperative neurological deficits. Three patients had neuropathic pain, which settled with medication in the postoperative period. Five patients developed a surgical site occurrence. The mean duration of hospitalisation for these patients was 7.7 days. The mean follow-up period was 27.8 months. 90% (18) of the patients had classical schwannoma on histopathology.

Conclusion: Management of large presacral tumours is a daunting task even for the most experienced surgeons. Intracapsular excision of these tumours needs to be considered while planning these operations, based on the possible morbidity profile for these patients.

Keywords: Benign tumours, Giant schwannoma, Intracapsular excision, Pelvic tumours, Surgery

INTRODUCTION

Presacral tumours are an extremely rare entity which only occurs at a rate of one in every 40000 hospital admissions [1]. Schwannomas account for only 3% of tumours in this region [1]. These lesions are usually benign, unifocal and are often asymptomatic [2]. A few of these patients may present with compressive symptoms to the surrounding structures or occasionally with neurogenic pain [3].

Imaging plays an important role in the characterisation and preoperative evaluation of presacral masses. MRI is the investigation of choice due to its superior soft tissue resolution. It helps to identify key imaging features to make a diagnosis or narrow down the differential diagnosis, determine the anatomical extent of the lesion, plan image-guided tissue sampling if required, and aid in treatment and surgical planning. Schwannoma and neurofibroma are nerve sheath tumours that occur in the presacral region [4]. Though sometimes the two cannot be differentiated due to overlapping imaging findings, certain imaging features can point to a particular diagnosis. Preoperative histopathological confirmation may not be possible due to its location or inconclusive due to the large size of these tumours [5].

Surgical excision is the mainstay of management, although it is associated with high morbidity. The extent of the excision and the surgical approach must be tailor-made for each patient and vary primarily based on the tumour location, proximity to vital structures and the expertise available at the centre [2].

Although a few case reports are available, large datasets are sparse. The largest available series in literature is of seven patients in a multi-institutional series [2]. The present study included 20 patients with presacral schwannomas who underwent surgery in a Tertiary Care Centre in Tamil Nadu, India, with follow-up periods as long as 10 years.

MATERIALS AND METHODS

The present retrospective descriptive study was conducted in General Surgery Unit-IV, Christian Medical College and Hospital, Vellore, Tamil Nadu, India, from July 2013 to July 2024. The study proposal was approved by the Institutional Review Board. Data was collected retrospectively from the hospital database, and 20 patients were identified.

Inclusion and Exclusion criteria: Histologically/radiologically confirmed schwannoma patients (age of more than 18 years) with tumours located in the presacral region, with or without sacral

involvement, who underwent definitive surgical resection (anterior, posterior, or combined approaches) with complete medical records were included. Patients below the age of 18 and patients with non schwannomata presacral tumours were excluded. Presacral schwannomas that did not undergo surgical excision at our centre were also excluded.

The demographic profile, presenting symptoms, imaging findings, preoperative assessment, surgical details, postoperative complications, and follow-up were recorded.

STATISTICAL ANALYSIS

Data collected were analysed using Microsoft Excel (Microsoft 365 MSO Version 2505). Descriptive statistics were calculated, with mean and median serving as the measure of central tendency and the standard deviation representing the measure of dispersion.

RESULTS

A total of 20 patients were identified in eleven years. There was a male predominance of 13 patients. The most common symptom was urinary complaints because of the pressure effect [Table/Fig-1]. The imaging of choice was MRI, with 19 patients undergoing an MRI. The one patient who underwent a Computed Tomography (CT) scan had presented to the Emergency Department with a history of lower abdominal pain and had undergone evaluation for an acute abdomen. 18 patients had classical schwannoma on histopathology. One patient had a plexiform schwannoma, and the other patient had a schwannoma-neurofibroma.

Variables	Characteristics	Frequency (total=20)
Sex	Male	13
	Female	7
Symptoms	Urinary symptoms	9
	Neurogenic pain	5
	Bowel symptoms	3
	Abdominal pain	2
	Asymptomatic	4
Preoperative biopsy	Yes	17
	No	3
Level of lesion	Above S3	6
	Below S3	2
	Both above and below S3	12
Immediate postoperative complications	Nil	9
	CDG<3	9
	CDG>=3	2
Long-term complications		3
Follow-up	Lost to follow-up	5
	No recurrence	12
	Residual disease	3

[Table/Fig-1]: Patient demographics, and disease details (CDG- Clavien-Dindo Grading of postoperative complications).

Preoperative assessment: Seventeen patients had an image-guided core biopsy before surgery. Of these, biopsies of 16 patients were schwannomas, and the biopsy of one patient was suggestive of a neurofibroma. In the remaining patients, a biopsy could not be performed because of location; their imaging discussed in the multidisciplinary meeting was noted to have MRI findings suggestive of a benign nerve sheath tumour and hence underwent excision. One patient who had a preoperative biopsy of neurofibroma was found to have a schwannoma on postoperative biopsy. There were no other discrepancies noted in the preoperative and postoperative biopsies.

Surgical approaches: The authors observed that 19 patients underwent a laparotomy (transperitoneal approach) with intracapsular excision of the tumour, with the patient being positioned in the Lloyd-Davies position for ease of access into the pelvis. Only one patient required excision of the surrounding viscera. In this patient, there was a giant presacral tumour with the sigmoid mesocolon and mesorectum densely adherent to it. The patient underwent an en-bloc excision of the tumour along with a rectosigmoidectomy and primary anastomosis with a stapling device.

The mean operating time was 2.85 hours (±1.12), with a range of 1.5 hours to 5.5 hours. Blood loss during the operations varied from 100 mL to 900 mL with a mean blood loss of approximately 500±240 mL, with the exclusion of one patient who suffered inadvertent injuries to the common iliac artery bifurcation and the external iliac vein at the time of excision, requiring a damage control surgery with packing of the presacral region, followed by a re-exploration after 24 hours. The total estimated blood loss for that operation was about 7 litres [Table/Fig-2].

Case no.	Age (years)	Size of the lesion (cm)	Blood loss (mL)	Hospital stay (days)	Follow-up (months)
1	43	11×10×9.5	NA	7	12
2	56	13.2×11.3×10	700	7	72
3	31	12.1×11.6×11.8	500	6	126
4	21	12.5×10×11.7	300	7	10
5	50	17×10×14	NA	6	96
6	40	9.5×10×14.5	900	9	6
7	40	11.5×11×10.5	750	8	3
8	24	8.9×10.6×6.6	800	15	8
9	42	9.3×8.3×9	700	7	2
10	25	8×7.5×9	500	3	16
11	40	7.1×9×8.8	450	8	1
12	38	6.8×6.8×4.2	150	7	48
13	49	14.7×14×12.4	7000	15	1
14	38	8.7×6.5×6.5	300	10	32
15	34	25×16.4×15	400	5	1
16	72	9.8×8.3×9.7	350	8	1
17	65	8×8×10.3	NA	6	2
18	58	9×8.6×9.3	300	8	12
19	41	7.4×7.8×10.4	900	7	1
20	20	7×5×7	100	6	1

[Table/Fig-2]: Operative details, hospital stay and duration of follow-up.

Postoperative complications: The mean duration of hospitalisation for these patients was 7.7 days (±2.9) with a range of 3-15 days. Two patients developed postoperative neurological deficits. One of these patients had a transient sensory loss, which settled during his stay in the hospital. One gentleman developed both sensory and motor deficits, which required musculoskeletal physiotherapy. Three patients had neuropathic pain, which settled with medication in the postoperative period, and they were asymptomatic at the time of follow-up.

Five site infection were there which required laying open of the wound bedside and daily dressings. One patient developed a wound dehiscence requiring a re-laparotomy and closure of the rectus.

Two patients developed postoperative urinary retention following removal of the urinary catheter; however, on follow-up, they underwent catheter removal and did not have further urinary symptoms. Two patients developed a urinary tract infection in the postoperative period. One patient had a small pelvic collection which could not be drained by pigtail placement and was managed with antibiotics [Table/Fig-3].

On long-term follow-up, two patients developed incisional hernias and underwent mesh repair for the same. One patient developed adhesive intestinal obstruction in the follow-up period and was managed non-operatively [Table/Fig-3].

Postoperative complications (CDG<3)	Frequency
Surgical site infection	4
Postoperative urinary retention	2
Urinary tract infection	2
Pelvic collection	1
Long-term complications	
Incisional hernia	2
Adhesive intestinal obstruction	1

[Table/Fig-3]: Distribution of postoperative complications (CDG<3) and incidence of long-term complications.

Follow-up: Four patients were lost to follow-up. Of the remaining patients, the mean follow-up period was 27.8 months. One patient had a recurrence of disease, review of pathology showed schwannoma-neurofibroma, on further imaging had multi-focal disease and was managed symptomatically.

DISCUSSION

Schwannomas are one of the most common of peripheral nerve sheath tumours. Most of these tumours are asymptomatic when they are small. As they progress in size, patients may develop symptoms either due to compression of adjacent structures or neuropathic pain due to compression of nerve root [6]. Leclerc A et al., in their series of six patients describe that about 50% patients presented with radicular pain and only about 16% patients presented with compressive symptoms, i.e., constipation [7].

Giant presacral tumours were defined as those tumours having >50 mm diameter on cross-sectional imaging [7]. The average size at the time of presentation in the present case series was about 11.63 cm, with all the cases with the least diameter more than 5 cm.

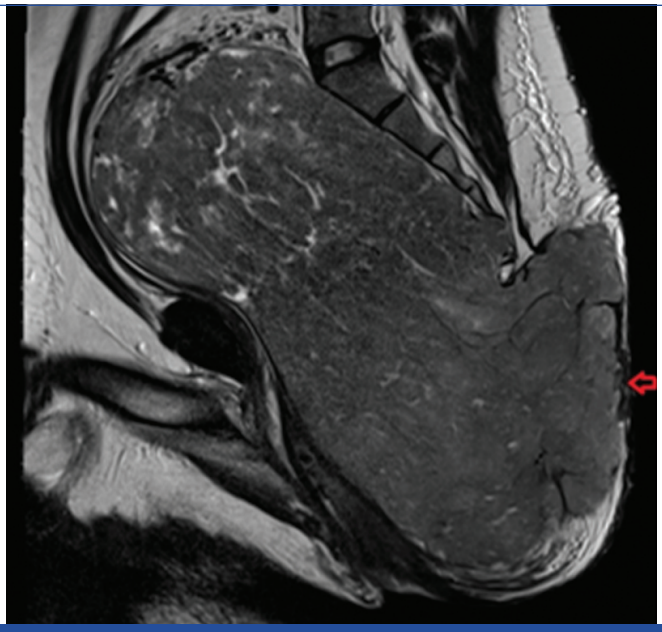
Although CT may be able to give us the extent of the lesion, MRI still offers an advantage in pelvic masses and help us delineate the mass from the surrounding structures, in addition to giving us information regarding infiltration into the neural foramina, which is vital in these cases as per Pennington Z et al., [2].

MRI is the modality of choice in the evaluation of presacral nerve sheath tumours [8]. On MRI, presacral schwannomas are large well encapsulated lesions with T2W heterogenous high signal intensity with cystic changes and low signal intensity on T1 weighted images. A thickened nerve root may be seen with extension into and widening of neural foramina [4]. Large lesions cause remodelling of the sacrum and not destruction. Neurofibromas show a characteristic 'target sign' on MRI with central T2W hypointensity due to fibrous core and peripheral T2W hyperintense rim of myxoid material [4]. All the patients underwent an MRI except the one patient who presented to the Emergency Department and underwent to Contrast Enhanced Computed Tomography (CECT) abdomen and pelvis.

A routine preoperative biopsy is not required in many cases of presacral schwannoma if typical imaging characteristics are present. In presacral masses, if the lesion is inoperable, if there is a strong suspicion of malignancy, suspicion of lymphoma, or those lesions which cannot be adequately characterised on imaging, may be subject to preoperative biopsy [9]. Few authors suggest preoperative biopsies for presacral tumours only if they would significantly alter patient outcomes [10]. While some authors advocate preoperative biopsies as long as it is anatomically feasible [11]. The authors followed a protocol of doing preoperative biopsy whenever anatomically feasible, so the authors did a biopsy in 17 out of 20 patients.

Image-guided core needle biopsies were done for most of the patients; however, as suggested by Debaibi M et al., MRI may be used as a gold standard for diagnosis and avoid preoperative biopsies- which may have potential complications of haemorrhage or tumour seeding in the case of a malignant tumour [12].

Surgical approach to these tumours can be decided based on the Klimo classification with a combined anterior-posterior approach for Type II tumours (tumours that have eroded into the sacrum and transperitoneal (anterior) approach for Type III tumours (tumours confined to the presacral space). All the patients were Klimo type III. As suggested by Klimo P et al., 19 out of the 20 patients underwent an anterior approach [13]. One of the patients underwent a perineal approach for excision, in the prone jack-knife position. The tumour in this patient was large (25×16.4×15 cm) but was confined to below the S3 level extending on the subcutaneous plane and skin ulceration in the perineum [Table/Fig-4]. The authors continued to opt for the anterior approach for all the other patients. In a previous study the authors preferred the anterior approach for most of their Klimo type III patients and posterior approach in the patient with a mass which was Klimo type II. If the tumour was large enough to extend into the abdominal cavity, the posterior approach is not feasible [14].

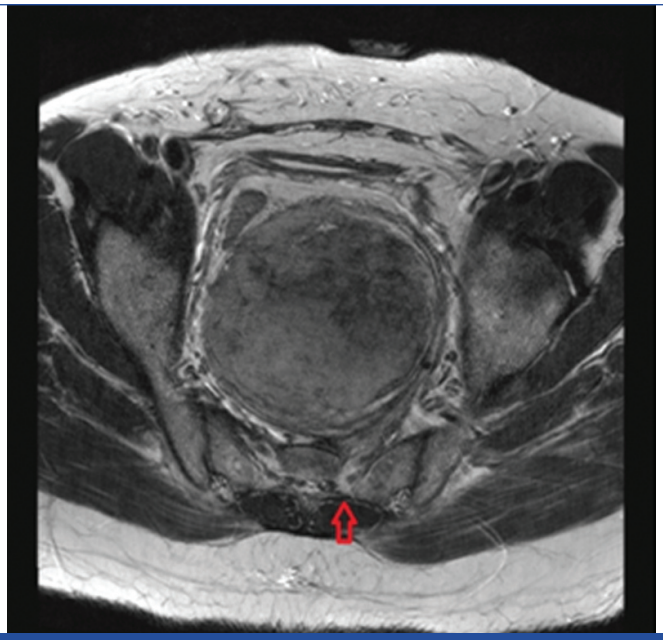


[Table/Fig-4]: Sagittal cuts from T2-weighted MRI pelvis extending from the S3 level to the subcutaneous tissue in the perineum with focal ulceration on the skin (arrow).

Complete excision of these tumours remains the mainstay in management [15]. However, due to the location of these tumours and proximity to vital structures, this may not be feasible. At our centre, we have opted for intracapsular excision of these tumours. Even with the three patients with residual tumours, follow-up with 6-monthly imaging shows negligible increase in size, with no recurrence of symptoms. The three patients with residual disease were the ones who had lesions extending into the foramen and no preoperative neurogenic symptoms [Table/Fig-5].

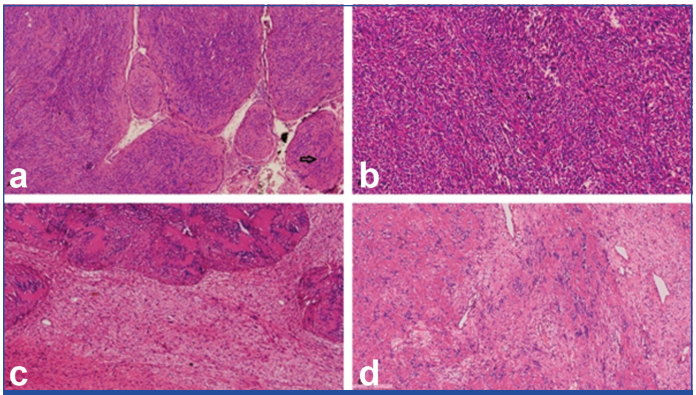
Complications occur frequently following complete excision of these tumours, up to 33%, although transient [16]. About seven of the patients had surgery-related complications, which were comparable to other studies [1,6,10,13,17,18]. According to Pennington Z et al., in their series of seven patients, the mean blood loss was about 1.7 litres. In the present series, mean blood loss was about 890 mL [2]. The mean operating time is usually about 4 hours in giant presacral tumours with two skilled operators [7]. The mean operating time in present cases was about 2.85 hours.

According to Leclerc A et al., average duration of stay was about 6.16 days in comparison to the mean duration of hospital stay, which was about 7.7 days. The average duration of follow-up in these patients was 27.8 months, which was significantly less than literature, which is about 67 months [7]. The authors attribute this to the fact that most of the patients were from faraway places which lead to infrequent follow-up visits.

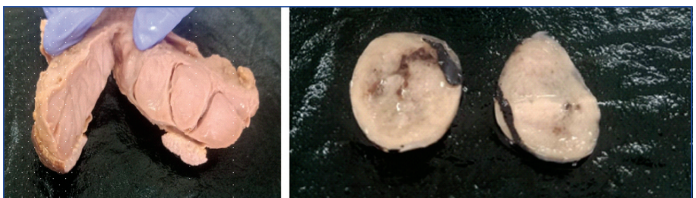


[Table/Fig-5]: Axial section of T2-weighted MRI showing mild tumour extension (arrow) into the S2 neural foramen.

Three patients had a residual lesion which, on follow-up, did not show any increase in size and hence did not require reoperation. Conti et al., report recurrence rates of about 4.5% [19]. Treatment of these recurrences was dependent on their site and symptomatology. There was only one recurrence of disease for which the patient was kept on follow-up. The histopathology for the patient with recurrent disease was schwannoma-neurofibroma, which belongs to benign Hybrid Peripheral Nerve Sheath Tumours (HPNST) [Table/Fig-6], while the others were predominantly schwannomas [Table/Fig-7]. Further planning based on the pathology also plays a key role in the follow-up of these patients. These tumours are known to be associated with multifocal presentations, and it may be prudent to screen for other foci. This patient was found to have multiple small lesions along the lower thoracic and lumbar spine, discussed in the multidisciplinary team meeting and kept on follow-up. No set protocols were available in the literature for the management of recurrences, and they were managed on a case-by-case basis based on symptomatology.



[Table/Fig-6]: a) (Eosin-haematoxilin - 100x): Multinodular cellular tumour with benign spindle cells in sheets and fascicles with focal nuclear palisading (indicated by arrow); b) (100x): Hypercellular tumour composed of interlacing fascicles and bundles of spindle cells and focal herring bone pattern; c) (40x): Schwannian nodules with Verocay bodies alternating with loose neurofibroma component; d) (10x): Cellular areas Antony A with nuclear palisading (Verocay bodies) and a hypocellular component (Antony B).



[Table/Fig-7]: (Left) Plexiform schwannoma - Multinodular soft tissue tumour with homogenous, soft to firm glistening cut surface, (Right) Schwannoma - Circumscribed mass with a tan-grey white to myxoid cut surface with cystic and haemorrhagic areas.

Limitation(s)

Due to the rarity of presacral schwannomas, the size of the study is limited, which reduces the statistical power of the analysis. As presacral schwannomas are often asymptomatic and incidentally detected, the paper may have disproportionately included symptomatic or surgically treated cases, potentially skewing the clinical and radiological characteristics observed. The present study is retrospective. Further multicentric prospective studies with larger patient cohorts are warranted to validate these findings and guide evidence-based management.

CONCLUSION(S)

Management of large presacral tumours is a daunting task even for the most experienced surgeons. Adequate preparation with appropriate preoperative cross-sectional imaging and biopsy is vital in surgical planning. Choosing the right surgical approach, tailormade to each patient, is by far the most important task in reducing complications for these patients. The authors recommend preoperative MRI for assessment, and if the MRI shows classical features of a schwannoma, preoperative biopsy may be avoided. Transperitoneal approach is recommended as a preferred approach. Regular follow-up of patients with yearly imaging studies is advisable.

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PLAGIARISM CHECKING METHODS: [Jain H et al.]
- Plagiarism X-checker: Mar 12, 2025
- Manual Googling: Jul 08, 2025
- iThenticate Software: Jul 10, 2025 (5%)

ETYMOLOGY: Author Origin

EMENDATIONS: 6

AUTHOR DECLARATION:
- Financial or Other Competing Interests: None
- Was Ethics Committee Approval obtained for this study? Yes
- Was informed consent obtained from the subjects involved in the study? Yes
- For any images presented appropriate consent has been obtained from the subjects. Yes

Date of Submission: **Mar 02, 2025**
Date of Peer Review: **May 21, 2025**
Date of Acceptance: **Jul 12, 2025**
Date of Publishing: **Apr 01, 2026**