

CASE REPORT

Retroperitoneal ancient schwannoma presenting as left flank pain and moderate unilateral hydronephrosis: A case report and literature review

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Key Clinical Message

Benign ancient retroperitoneal schwannomas (BARS) exhibit abdominal masses and flank pain to incidental findings at more advanced stages. Histopathological and immunohistochemical analysis is essential for confirmation of benign nature. Our patient was misdiagnosed as ureteric colic, highlighting the need to consider BARS in differential diagnosis to prevent complications like hydronephrosis.

Abstract

Ancient schwannomas are usually benign neoplasms that originate from Schwann cells of peripheral nerves. We present a novel case of a 24-year-old young male with left flank pain and nausea which was initially thought to be left ureteric colic. However, in-depth imaging and biopsy revealed a retroperitoneal mass. The definitive diagnosis was narrowed down to Benign Retroperitoneal Ancient Schwannoma (BARS) via immunohistochemistry and histopathological analysis. This often marble-shaped S100 protein-positive tumor is an under-recognized and potential cause of hydronephrosis if localized near the renal structures. In addition, the retroperitoneal location with infrarenal abdominal aortic adherence is another rare peculiarity in the present case that demands prompt diagnosis and surgical excision to avoid any cardiovascular sequelae such as hypotension and abdominal pain, as indicated by the natural history of growth of this benign tumor. Therefore, timely excision of this benign tumor prior to its further proliferation is paramount. We initially planned laparoscopic removal but adopted excision via laparotomy because of the proximity of the vital structures. The post-operative course of the patient was uneventful and subsequently the patient's presenting complaint of left abdominal flank pain greatly improved. The patient was advised to undergo follow-up computed tomography scan of kidney ureter bladder and RFT evaluation 6 months postsurgery which indicated no evidence of recurrence or iatrogenic complications. The diagnosis and management of the present case share valuable experiences for similar future cases worldwide.

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KEYWORDS

ancient schwannoma, atypical left flank pain, benign tumor, case report, hydronephrosis, retroperitoneal mass

1 | INTRODUCTION

Benign nerve sheath tumors, such as benign ancient retroperitoneal schwannomas (BARS), are relatively rare entities that may remain asymptomatic until they grow enough to impinge on organ structures. A Japanese asymptomatic cohort had an average growth rate of 1.9 mm/year.¹ Only 0.7% of ancient schwannomas are localized in the retroperitoneum.² Initially, these masses may raise apprehensions of a malignant tumor on imaging and preoperative pathological diagnosis.³ Therefore, postoperative histopathological analysis of the excised mass is vital for confirming the benign nature of this neurilemmoma. Histopathology reveals hypercellular Antoni A regions containing spindle-shaped cells and Hypocellular Antoni B regions.⁴ Immunohistochemical staining of the S-100 protein is a frequent finding.⁵ Complete surgical resection remains the gold standard for symptomatic cases; however, there have been instances of utilizing radiation therapy for giant invasive sacral schwannomas.^{1,6} Retroperitoneal schwannomas are uncommon tumors, constituting approximately 1%–5% of all retroperitoneal masses.⁷ Schwannomas in the retroperitoneal region represent 0.75%–2.6% of all schwannomas,^{8,9} with the majority occurring in the limbs, head, and neck.¹⁰ These tumors are typically solid, encapsulated, and originate in the paravertebral region.⁷ Only a few cases of large retroperitoneal tumors causing compression of adjacent structures have been reported in the literature. As the ureters and aortic bifurcation are located in the retroperitoneal space, they can be easily compressed by retroperitoneal lumbar tumors. Retroperitoneal tumors, especially those situated in the lumbar region, are highly prone to precipitate hydronephrosis despite the mass size being less than 5 cm.⁶ Therefore, it is of great significance to promptly diagnose and surgically excise such masses to avoid various sequelae that would result later on from the tumor mass exerting pressure on adjacent soft tissues and other vital retroperitoneal structures such as flank pain, hydronephrosis, and external vascular occlusion. This article presents a rare case of retroperitoneal ancient schwannoma with compression of both the infrarenal aorta and the left ureter, resulting in moderate unilateral hydronephrosis. This case report has been reported in line with the CARE Criteria.

2 | CASE PRESENTATION

2.1 | Case History/Examination

A 24-year-old young male presented to the emergency department on the 1st of September, 2023, with a history of left flank pain for the past 10 days with complaints of nausea and 3 episodes of vomiting on the previous day. Before these 11 days, the patient attested to being in a normal state of health. The pain was sudden in onset and increased in intensity over the past 5 days. It was described by the patient as nonradiating, aggravated by lying down, not relieved by taking oral analgesics, and associated with vomiting. The vomit was described as nonprojectile, greenish in color, containing food particles, aggravated after meals, and relieved by antiemetics. The patient is a soldier by profession. His past medical and surgical history was not significant. Abdominal and testicular examination did not reveal any pathological findings. Abdominal examination on inspection showed no abnormalities of the skin, abdominal shape, movement, or any abdominal mass. Auscultation of abdomen did not show any bruits or rubs. On Palpation of the abdomen shifting dullness and fluid thrill were absent. On palpation of the right flank the patient showed no discomfort. However, the left flank palpation resulted in the patient complaining of significant tenderness. The pain was non radiating in nature and did not refer to another region. The flanks were symmetric and showed no discoloration. Baseline patient investigations were not significant and the patient was afebrile.

2.2 | Imaging Investigation Findings

Initially, abdominal ultrasonography and chest radiography were ordered by the physician, which revealed no abnormal findings. Plain radiography has limited use in detecting soft tissue tumors. However, they can be useful to exclude any bony involvement, which this case had none of. Ultrasonography may not be able to detect such masses occasionally due to their deep location's inside the human body leading to ultrasonic beam distortion. Other factors can be not knowing what to look for and an inexperienced sonographer. However, on an ultrasonogram of the kidney, ureter, and bladder (KUB), moderate left kidney hydronephrosis and left proximal ureteral dilation were observed.

The distal ureter was not visible because of abdominal gases. There was suspicion of an upper left ureteric calculus measuring 5.6 mm. The initial treatment plan was initiated on the lines of the suspected left ureteral colic. A CT-scan KUB ordered for ureteric calculi revealed an incidental finding of a 42.4×40.1×35.2 mm retroperitoneal mass. A Triphasic Computed tomography (CT) scan of the abdomen and pelvis confirmed this incidental finding. The mass was present to the left of the infrarenal aorta with loss of interface, and it was underneath the renal hilum, pushing the renal vessels superiorly, and was near the left ureter (Figure 1). It was decided to further investigate via a CT-guided tru-cut biopsy. Two fragile cores of 10 and 7 mm, along with two tissue core fragments measuring 2×2 mm, were fixed with 10% buffered formalin solution. The histopathologic report described tissue cores showing proliferation of spindle cells with fibrillary stroma and few admixed skeletal muscle fibers. No pleomorphism, mitotic activity, or necrosis was observed. Immunohistochemical staining revealed SRY-Box transcription factor 10 (+) positivity for S100 protein, Smooth muscle antibody (–), Desmin (–), and discovered on gastrointestinal stromal tumors 1 (–). A provisional diagnosis of a nerve sheath tumor without atypia and necrosis was adopted. Surgical excision was recommended for further evaluation and confirmation.

2.3 | Surgical Management

Initially, laparoscopy was planned by an on-call general surgeon. However, laparoscopic removal was avoided because of the close interaction of the schwannoma with the infrarenal aorta and the left renal vessels. Iatrogenic injuries, proper view of the surgical field, were the main factor behind choosing the method of excision. We did not want to risk any accidental ligation, laceration, kinking by suturing of the abdominal aorta, left ureter, or any other

nervous or vascular structure while ligating the feeding arteries of the schwannoma or while excising the mass itself. The patient and his family were explained the rationale of his surgical management plan and was onboarded with informed and written consent. A repeat pre-operative computed tomography scan of kidney ureter bladder (CT-KUB) showed no noticeable changes in the mass shape, size, and positioning in the left retroperitoneum. A laparotomy was conducted via a vertical midline incision extending from the epigastrium to 3 cm below the umbilicus by cutting through skin, subcutaneous tissue, and rectus sheath. The abdominal cavity was grossly examined for any peculiarities. This was followed by mobilization of the left transverse and descending colon using the Maddox procedure. Visual localization of the retroperitoneal schwannoma indicated it to be pushing the renal arteries superiorly, infrarenal aorta towards the midline as well as being in close relation to the left ureter. Following the localization of the tumor, feeding vessels of the tumor were ligated, and starting inferiorly, the mass was mobilized gradually and completely removed (Figure 2). The patient was closely monitored in the surgical ICU for 4 days post-operatively. Thereafter, he was shifted to the general surgery ward for 2 days and discharged.

Postsurgical excision of the neoplasm was performed for further histopathological analysis. Serial slicing of the specimen revealed a hemorrhagic cut surface. Multiple samples were collected in three blocks: A, B, and C. Histological sections of the tumor under microscopy revealed an encapsulated neoplasm composed of cytologically bland spindle cells with degenerative nuclei at certain places arranged in short fascicles containing more densely cellular Antoni A areas with nuclear palisading alternating with paucicellular Antoni B regions. Histopathological examination revealed degenerative atypia (Figure 3). The final diagnosis of BARS is based on preoperative and post-operative investigative evidence.

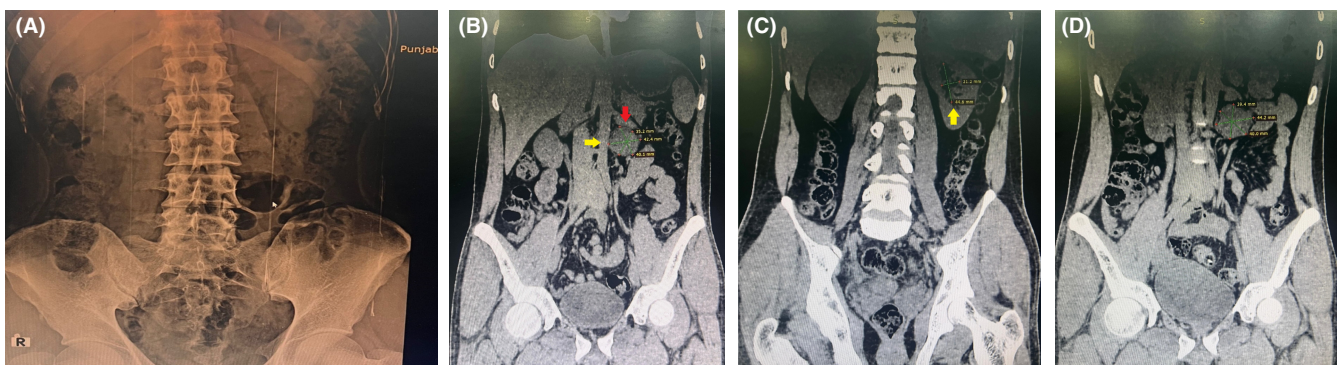


FIGURE 1 (A) X-Ray KUB showed no signs of any soft tissue mass. (B) Triphasic Abdominal CT displaying a 42.4×40.1×35.2 mm round, encapsulated, well circumscribed, homogenous mass in the left retroperitoneum impinging infrarenal aorta towards midline (yellow arrow), Left Renal artery superiorly (red arrow). (C) CT-KUB suggestive of Mild left sided hydronephrosis (yellow arrow), likely due to retroperitoneal mass impinging on the left ureter. (D) CT-KUB showing a well-defined encapsulated mass in close relation to vascular structures.

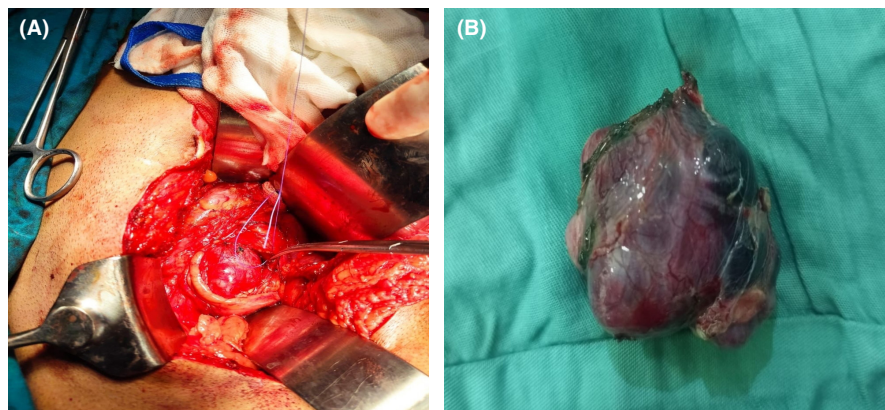


FIGURE 2 (A) Intraoperative view. (B) Gross appearance revealed a single encapsulated highly vascularized reddish-brown mass with a diameter measuring 45 mm.

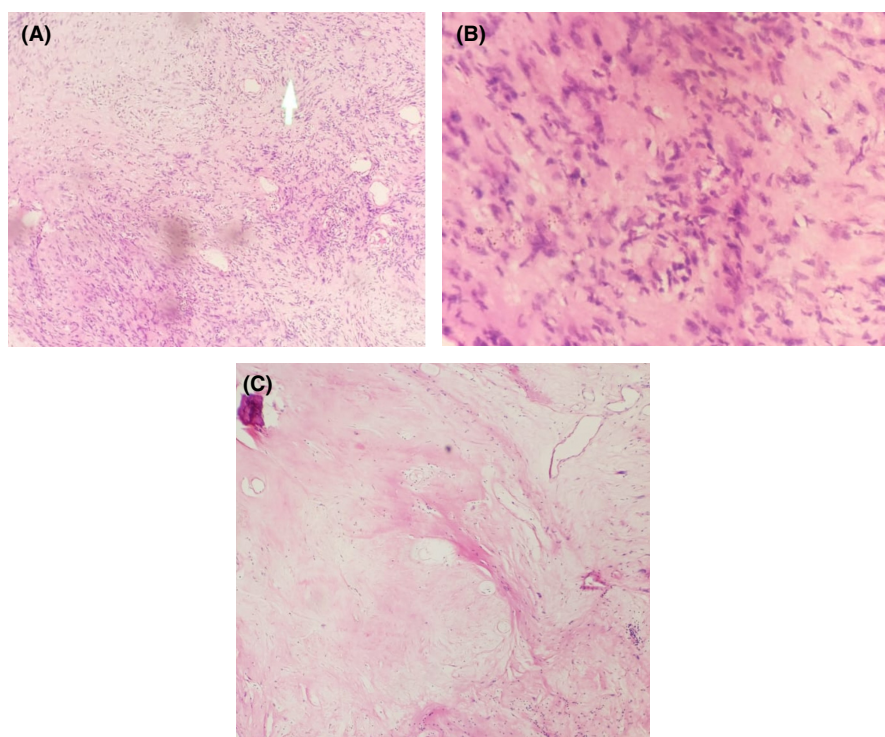


FIGURE 3 (A) Low-power view showing a predominantly cytologically bland spindle cell tumor with fibrillary stroma and few admixed skeletal muscle fibers (H&E, $\times 100$). (B) High-power view showing spindle cells in hypercellular areas which express Antoni A fibers. (H&E, $\times 400$). (C) High-power view showing hypocellular area expressing Antoni B Fibers (H&E, $\times 400$).

2.4 | Ongoing Follow-up Plan

The patient was regularly monitored for potential recurrences. The patient was advised to undergo follow-up CT-KUB and RFT evaluation 6 months postsurgery. The postoperative course of the patient was uneventful, and the left abdominal flank pain greatly improved. At the 6-month follow-up, no evidence of recurrence or any iatrogenic complication was observed.

3 | DISCUSSION

Schwannomas presenting with degenerative changes, such as cyst formation, hemorrhage, calcification, and hyalinization, are classified as a subtype of retroperitoneal schwannoma, namely, ancient retroperitoneal schwannoma.⁷ Macroscopically, schwannomas are

solitary, firm, well-circumscribed tumors with a smooth surface.¹⁰ Histologically, retroperitoneal schwannomas are distinguished by alternating Antoni A and B areas. Antoni A areas contain clusters of compact, elongated bipolar spindle cells arranged in a palisading pattern, whereas Antoni B areas are characterized by loosely arranged cells in a myxoid background.¹¹ However, large retroperitoneal and pelvic schwannomas have been reported to exhibit uniform spindle cell morphology without distinct Antoni A and Antoni B areas.¹² Immunohistochemically, positive staining for S-100 protein in the cytoplasm of schwannoma cells is a reliable aid for accurately diagnosing these tumors.¹³ Our immunohistochemical staining results are in agreement with this statement. Malignant retroperitoneal schwannomas can be identified by abnormal histological features, such as excessive blood vessel infiltration, mitotic figures, and nuclear atypia.¹⁴

The high flexibility of the retroperitoneal space often results in retroperitoneal schwannomas presenting without specific symptoms in the early stages. However, as the stromal, extracellular components, and benign tumor cells proliferate, the tumor stiffens and invades adjacent normal tissue and empty retroperitoneal spaces.²¹ Eventually, the tumor becomes stiffer than the surrounding softer tissue and displaces it. Allowing for further tumor invasion as well as generation of significant mechanical forces which eventually result in the compression symptoms associated with vascular, lymphatic, nervous, and renal structures.²¹ This is the general pathophysiological mechanism behind all cases in (Table 1). Manifestations can range from an abdominal mass and flank pain to incidental findings at more advanced stages, leading to delayed diagnosis and treatment.¹⁵ A study from Singapore reported that the symptoms were nonspecific, with neurological symptoms being rare.⁷ Common symptoms include vague abdominal pain, flank pain, hematuria, headache, secondary hypertension, and recurrent renal colic pain.¹⁰ In this study, a 24-year-old male patient presented with atypical symptoms including nonradiating left flank pain, worsened by lying down, not relieved by oral analgesics, and accompanied by nonprojectile vomiting. Imaging investigations also revealed a nonspecific, large retroperitoneal mass complicated by moderate unilateral hydronephrosis, highlighting the importance of considering a differential diagnosis of ancient schwannomas during initial evaluations by clinicians. Table 1 provides a detailed description of other symptomatic cases of BARS leading to varying degrees of hydronephrosis.

As the preoperative diagnosis of retroperitoneal schwannomas is challenging owing to the lack of pathognomonic features, imaging investigations such as ultrasonography, CT scans, and magnetic resonance imaging (MRI) are crucial during the initial evaluation of the tumors. Ultrasonography is both useful and cost-effective for tumor detection. CT scans can identify well-defined areas of low or mixed attenuation with cystic necrotic centers, with cystic changes being more common in retroperitoneal schwannomas than in other retroperitoneal tumors.¹⁰ MRI provides superior visualization of large retroperitoneal tumors, including their origin, vascular architecture, and involvement with other organs.⁷ CT-guided biopsy and fine-needle aspiration are reliable diagnostic tools for retroperitoneal schwannomas, particularly when the biopsy sample contains sufficient Schwann cells for microscopic analysis.¹⁰

The differential diagnoses for schwannomas include fibrosarcoma, liposarcoma, and ganglioneuroma, which exhibit similar findings on CT and MRI.¹⁶ Preoperative misdiagnosis of retroperitoneal schwannomas are relatively common, underscoring the importance of ultrasound-guided biopsy. However, cellular

pleomorphism in degenerated areas can lead to inaccurate diagnosis, although biopsy remains useful for suspected malignant lesions.¹⁷ Additionally, many researchers advise against preoperative biopsy due to the risks of hemorrhage, infection, and tumor seeding.^{15,18} In this case, CT-guided tru-cut biopsy of the mass revealed proliferation of spindle cells with fibrillary stroma, along with a few admixed skeletal muscle fibers, without evidence of pleomorphism, mitotic activity, or necrosis, consistent with the postoperative biopsy examination. Consequently, a provisional diagnosis of a nerve sheath tumor without atypia and necrosis was adopted. Given the inability to definitively rule out malignancy preoperatively due to limitations in accurate pathological diagnosis, complete surgical excision with negative soft resection margins is recommended for managing retroperitoneal schwannomas.¹⁸ However, there is debate regarding the necessity of excising adjacent nonneoplastic tissues and viscera. Literature suggests that simple enucleation or partial excision without unnecessarily harming adjacent tissue may suffice due to the benign nature of most cases.^{10,15}

The prognosis of BARS is highly favorable, with rare reports of recurrence; however, close monitoring is essential following their removal. Of the cases in Table 1 only Ragurajprakash et al. had a reoccurrence of the tumor that too because of its initial partial resection years ago. It again underwent a subtotal resection due to its stiffness with the use of radiation therapy for the remaining tumor mass. There were no further neurological deficits on follow-up in this case.⁶ All other cases with available data were simple laparotomies or laparoscopies with no recurrence on follow-up, indicating a good outcome. Surgical resection is advised in cases of recurrence, as retroperitoneal schwannomas typically exhibit low sensitivity to radiation and chemotherapy, rendering adjuvant therapy ineffective.¹⁸ In recent years, laparoscopic excision¹⁹ and robotic resection²⁰ have emerged as promising surgical techniques. Nevertheless, the choice of surgical approach may be influenced by factors such as tumor location and size. In this case, laparotomy was performed via a vertical midline incision extending from the epigastrium to 3 cm below the umbilicus by cutting through the skin, subcutaneous tissue, and rectus sheath. The abdominal cavity was grossly examined for any peculiarities. Subsequently, the retroperitoneal schwannoma was completely excised following visual localization, which showed displacement of the renal arteries superiorly, infrarenal aorta towards the midline, and proximity to the left ureter. It was decided to regularly monitor the patient for any potential recurrence.

Our patient was advised a follow-up CT-KUB and renal function evaluation at 6 months postsurgery. Ongoing follow-up is required in cases of postoperative recurrence or other complications, including renal failure. In general, as

TABLE 1 Literature review for Cases of BARS associated with hydronephrosis.

Author (year)	Age	Sex	Symptoms	Location	Initial Diagnosis	Diagnostic Aids Used	Excision Method	Recurrence
Blum et al. (1985)	21	Female	Seizures	Left pararenal mass arising from paraspinous ganglia	NFI	CT, IVP, HP	Laparotomy	—
Cortés et al. (1999)	42	Female	—	Left Uretero-pelvic left junction	—	Abdominal USG	Laparotomy	—
Maeda et al. (2000) ^a	36	Male	Difficult urination	Retovesical space	—	IVP, CT, MRI, HP	—	—
Girgin et al. (2003)	73	Male	Bilateral hydronephrosis	Abdomino-pelvic mass extending from epigastrium to pubic symphysis.	Pelvic mass since last 4 years	USG, CT, HP	Laparotomy	No recurrence at 6 months
Matsuo et al. (2006)	63	Male	Left Ureter stone and left hydronephrosis	Left Retroperitoneum arising from femoral nerve	—	USG, CT, MRI	Microscopic Surgery	No evidence
Curry et al. (2007)	59 26 68	Male Female Male	UTI LFP UTI	Left or right reno-adrenal topography	Left adrenal tumor and UTI	USG, MRI, HP	Laparotomy	No evidence
Ozbird et al. (2011)	73	Male	Swelling, vague abdominal pain, and abdominal mass	Epigastrium to symphysis pubis	Retro-peritoneal tumor	USG, CT, HP	Anterior Laparotomy	No recurrence at 9th year follow-up
Ozbird et al. (2011)	46	Female	Flank pain, one episode of rectal hemorrhage, and increasing edema at the lower extremities	Right kidney lower pole to the pelvis	Retro-peritoneal tumor	USG, CT, HP	Anterior Laparotomy	No evidence of recurrence in the last 28 months of follow-up.
Zhang et al. (2018)	34	Female	Left abdominal pain for 1 month	In Close relation to the left ureter	Retro-peritoneal tumor	CT, Pyelography, HP	Laparoscopic removal converted to laparotomy	No evidence of recurrence on the 12-month follow-up.
Ragurajaprakash et al. (2020)	56	Female	Renal Failure due to hydronephrosis	Invasive in retro-peritoneum	Recurring Giant Sacral schwannoma	CT angiogram, CT, MRI	Laparotomy (subtotal excision) & radiation therapy	Regularly followed up without neurologic deficits.
Our Case (2024)	24	Male	LFP, Vomiting	Left RP in close relation to AA aorta, Left ureter and	Ureteric Obstruction due to potential calculus.	US-guided Biopsy, USG and CT.	Laparotomy	No evidence of recurrence or iatrogenic complications at 6 month follow-up.

Abbreviations: CT, Computed Tomography; HP, Histopathology; IVP, Intravenous Pyelogram; LFP, Left Flank Pain; MRI, Magnetic Resonance Imaging; NF-1, Von Recklinghausen's Neurofibromatosis; US/USG, Ultrasonography; UTI, Urinary Tract Infection.

^aThe right ureter and bladder were pushed to the left by the BARS but without the appearance of hydronephrosis on IVP.

described by (Table 1) BARS patients were checked for recurrence on follow-up for their presenting complaint such as neurological deficits, abdominal pain, and flank pain every 6 months. Some cases even extended their follow-up periods to 12 months and even beyond to 28 months in the case of Ozbir et al. (Table 1). Repeat imaging studies including CT scans were done to screen for a reoccurring mass due to a possible incomplete resection. A patient's outcome was deemed to be positive if no mass detected on follow-up.

4 | CONCLUSION

Our case demonstrates that the prognosis is favorable for total resection of BARS. Such rare tumors should always be included in the differential for retroperitoneal masses by clinicians. Prior awareness of such entities will lead to a quicker diagnosis and management of the neoplasm before it grows enough to exert pressure on surrounding tissues and structures. Future research in this area requires the development of tailor-made guidelines and scales according to the organ system affected, which could direct the treatment strategy towards wait and watch or surgical, and if surgical then whether laparoscopic or invasive laparotomy methods. Large-scale observational studies are needed to validate the benefit of a CT scan in asymptomatic patients, incidentally discovered via an ultrasound study due to the considerable radiation exposure risks. Although ancient schwannoma's do not respond well to radiation therapy, there has been an instance of its use in a recurring giant sacral schwannoma case management. In Neurofibromatosis Type 2-associated, merlin-deficient tumors various tyrosine kinase receptor inhibitors have shown promise. More pharmaceutical drug research should be launched in search of an effective chemotherapeutic drug for this benign tumor. Use of improved microsurgery technology may reduce recurrence rates.

AUTHOR CONTRIBUTIONS

Riyan Imtiaz Karamat: Conceptualization; supervision; validation; visualization; writing – original draft; writing – review and editing. **Ajeet Singh:** Project administration; supervision; validation; visualization; writing – original draft; writing – review and editing. **Adeel Anwaar:** Project administration; supervision; visualization; writing – original draft; writing – review and editing. **Zaka Ullah Malik:** Data curation; visualization; writing – original draft; writing – review and editing. **Javaid Hashmi:** Data curation; visualization; writing – original draft; writing – review and editing. **Muhammad Talha Haseeb:** Visualization; writing – original draft; writing – review and editing. **Aymar Akilimali:** Validation; visualization; writing – original draft.

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CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.


ETHICS STATEMENT

This is a case report utilizing anonymized patient information and so was classified as exempt from review from the Institutional Review Board.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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