



Primary bladder schwannoma: a case report and literature review

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Abstract: Primary bladder schwannoma is an extremely rare bladder tumor that originates from Schwann cells in the nerve sheath and often associated with von Reichenhausen's disease. Isolated cases of urinary bladder schwannoma are incredibly rare with no more than 1/1,000 of bladder tumours. We report a 33-year-old female patient who did not have any symptoms and was found by computed tomography (CT). Preoperative cystoscopy revealed a large sessile and smooth-surfaced mass on the anterior top of the bladder. Then she was successfully managed by partial cystectomy. Hematoxylin-eosin (HE) staining and immunohistochemistry (IHC) confirmed the mass was schwannoma. She was discharged 16 days after admission. In addition, she was followed up without intravesical recurrence or metastases for 29 months. Subsequently, literatures in PubMed (<https://pubmed.ncbi.nlm.nih.gov/>) accessed to bladder schwannoma since 1993 are searched and reviewed, more clinical data are provided to better assist in the diagnosis and treatment. In summary, bladder schwannoma is a rare benign tumor of the urinary system. Imaging examination and cystoscopy have a hint on the disease to a certain extent. The first choice of treatment is surgical resection, pathology is the gold standard and S-100 is usually positive. On account of the possibility of malignant transformation of the disease, Long-term follow-up is necessary.

Keywords: Bladder schwannoma; pathology and immunohistochemistry; case report

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Introduction

In the United States, a total of 81,400 new cases are increased with 17,980 deaths due to bladder cancer in 2020 (1). The most common bladder tumor is urothelial carcinoma. On the contrary, bladder interstitial tumor is relatively rare. Primary bladder schwannomas accounts for 0.1% of all bladder tumors (2). Schwannomas remain the most common ones in the head and neck (40–50%) (3), which could appear anywhere in the body where nerve sheath exists. However, schwannomas of the bladder are extremely infrequent. In the current study, we present the following article in accordance with the CARE reporting checklist (available at <https://dx.doi.org/10.21037/tcr-21-200>).

Case presentation

A 31-year-old female patient had a miscarriage due to scar pregnancy and a bladder mass was found by preoperative CT examination. One month after the operation, she came to our department. She did not show any lower urinary tract symptoms and hematuria. Her physical examination and laboratory data were also alright. Posterior flexible cystoscope revealed a 3.5 cm × 2.0 cm mass on the anterior top of the bladder (*Figure 1A*). Further computed tomography urography (CTU) was performed, demonstrating that the mass was enhanced, protruding to the bladder. The CT value of plain scan was 41 HU and the CT value of enhanced scan was 79 HU (*Figure 1B*). The patient has no basic diseases such as hypertension, coronary

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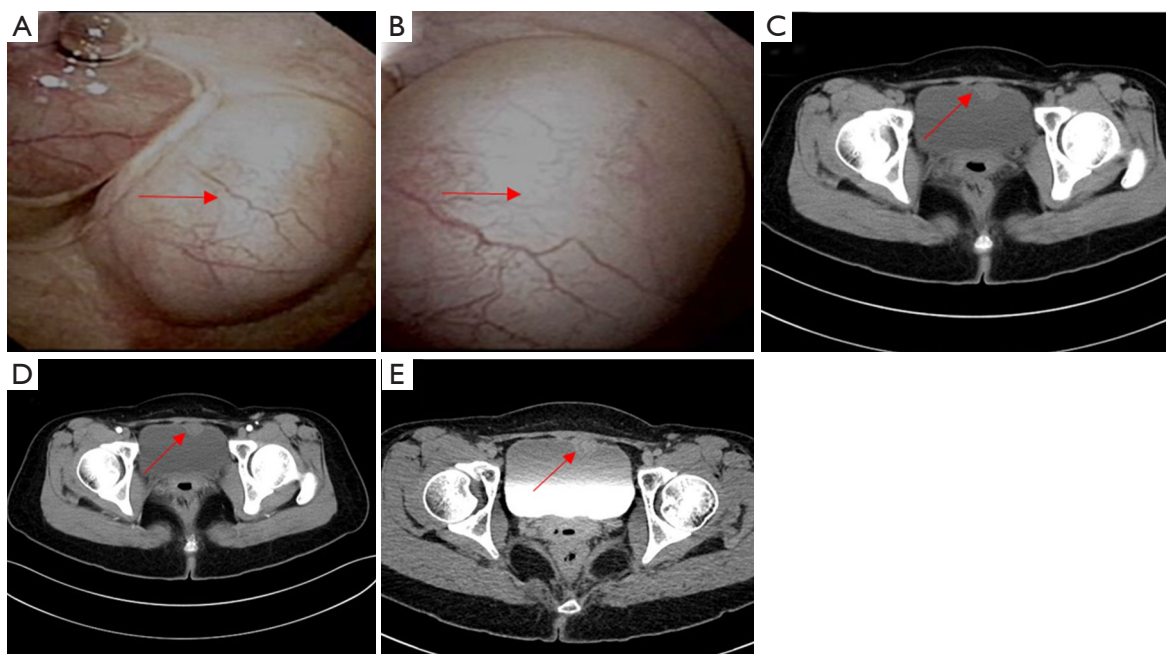


Figure 1 Preoperative examination. (A,B) Flexible cystoscope revealed a 3.5 cm × 2.0 cm mass on the anterior top of the bladder. (C) Non-contrast CT showed a slightly high-density mass with a CT value of 41 HU. (D) Enhanced CT demonstrated the mass was slightly enhanced with a CT value of 79. (E) The mass can also be seen in the excretory period. Arrows indicate the location of the tumor.

heart disease and diabetes with no family hereditary history and no mental illness. Preoperative CT evaluation did not find the evidence of metastasis in both lungs and abdominal organs. Then, the patient underwent a partial cystectomy. In the surgery, we chose a lower abdominal median incision to expose the bladder step by step. A tough mass could be observed on the anterior top when opening the bladder. The tumor was completely removed with a 1 cm safety margin. Intraoperative freezing indicates that the safety margin is negative and the section of the excised mass was incanus. The retropubic drainage tube and catheter were removed 7 and 8 days after the operation respectively. In addition, the patient achieved good recovery and was discharged one day later. Microscopically, the tumor cells were densely arranged in bunches and palisades, the nuclei were deeply stained and the local arrangement was loose (*Figure 2A,B*). Subsequent immunohistochemistry (IHC) was done which showed diffuse immunoreactivity for S-100 (*Figure 2C*), Vimentin was simultaneously positive (*Figure 2D*), but other markers including smooth muscle actin (SMA) (*Figure 2E*), Ki-67 was 8% (*Figure 2F*), desmin, cytokeratin, CD68 and beta-catenin were negative., the diagnosis of schwannoma was confirmed. After 29 months of postoperative follow-up, as indicated by cystoscopy and urinary color ultrasound

(*Figure 3A,B,C*), no metastases or recurrences were observed (*Figure 4*). All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient.

Discussion and review of the literature

Epidemiology and origin

The earliest case of schwannoma of the bladder was reported in 1993, which was presented as a pelvic mass (4). Schwannoma is a slow-growing myelinated nerve tumor that originates from Schwann's cells and is often associated with von Reichenhausen's disease. It usually occurs in the head and neck while bladder schwannoma is extremely infrequent and more common between the ages of 40 and 60 years (5). We searched the PubMed database. Up to 2020, there were 16 cases of bladder schwannoma. Given its rarity, we reported this case and reviewed the previous literature concerning bladder schwannoma (*Table 1*), including the reported years, regions, and clinical features.

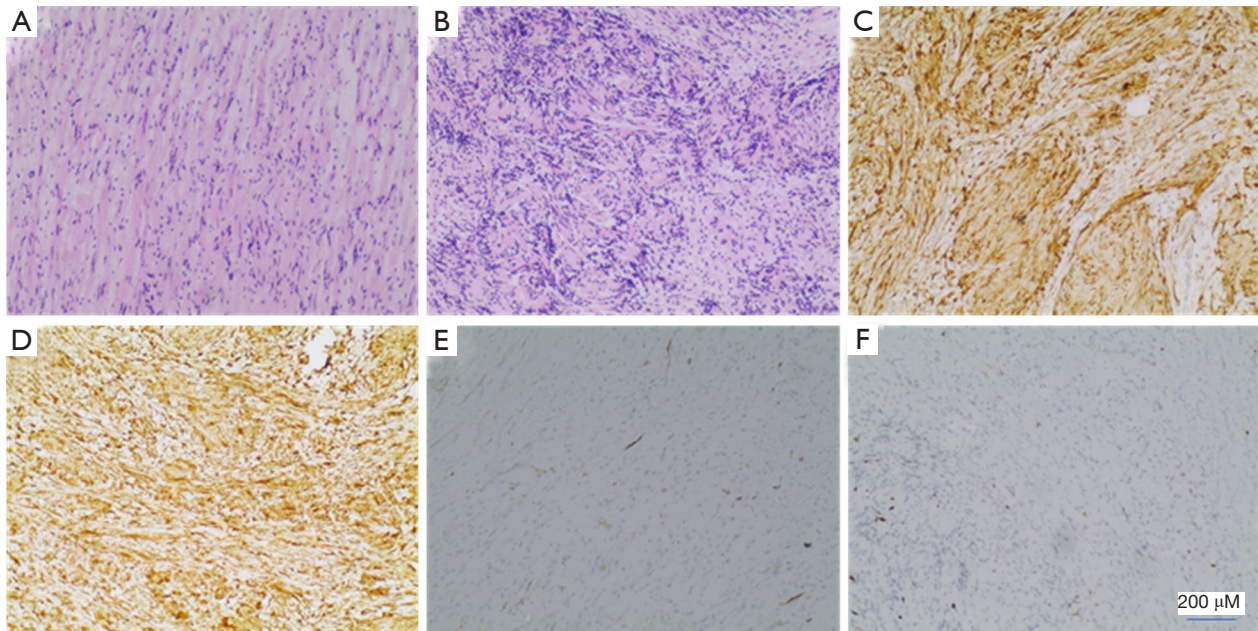


Figure 2 Histological findings of the tumor. (A,B) Hematoxylin and eosin staining indicating the tumor cells were densely arranged in bunches and palisades, the nuclei were deeply stained and the local arrangement was loose ($\times 200$). (C) Immunohistochemistry positive for S-100 ($\times 200$); (D) Positive for Vimentin ($\times 200$); (E) Negative for SMA ($\times 200$); (F) Positive for ki-67 (8%) ($\times 200$).

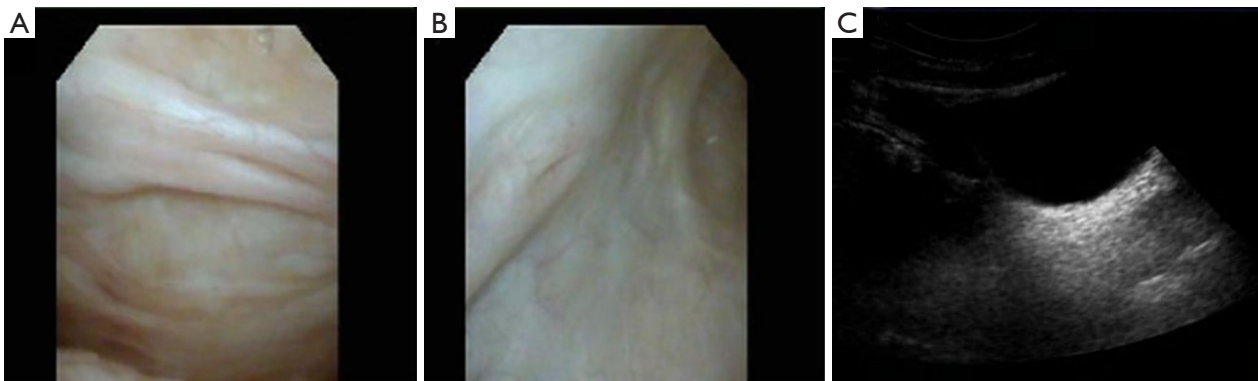


Figure 3 Postoperative reexamination. (A,B) Cystoscope showed postoperative scar (C) Urinary color ultrasound was normal.

Based on the above discussion, the most frequently reported cases were in the United Kingdom and the United States. With an average age of 49 and a median age of 50, the age of onset was between 25 and 88. There were 7 males and 10 females, revealing no significant gender difference.

Diagnosis

The most common clinical manifestations included lower urinary tract symptoms, for instance, pain, urgency and frequency (7/17, 41%), followed by hematuria (6/17, 35%),

Dyspareunia (1/17, 5%), vomiting (1/17, 5%) and recurrent infection (1/17, 5%). In addition, 5 cases were found accidentally. With an average of 4.4 cm, the size of the tumor ranged from 0.8 to 20 cm. Most tumors are found by ultrasound (US). US can be used as a bladder space-occupying screening due to its low price, convenience, and non-invasive characteristics. It is a pity that ultrasound was not performed in our case. Our case was detected by CT. CT showed single, intact, non-calcified and space-occupying lesions in the bladder area, showing isodensity or low density compared with muscle. Our case conforms

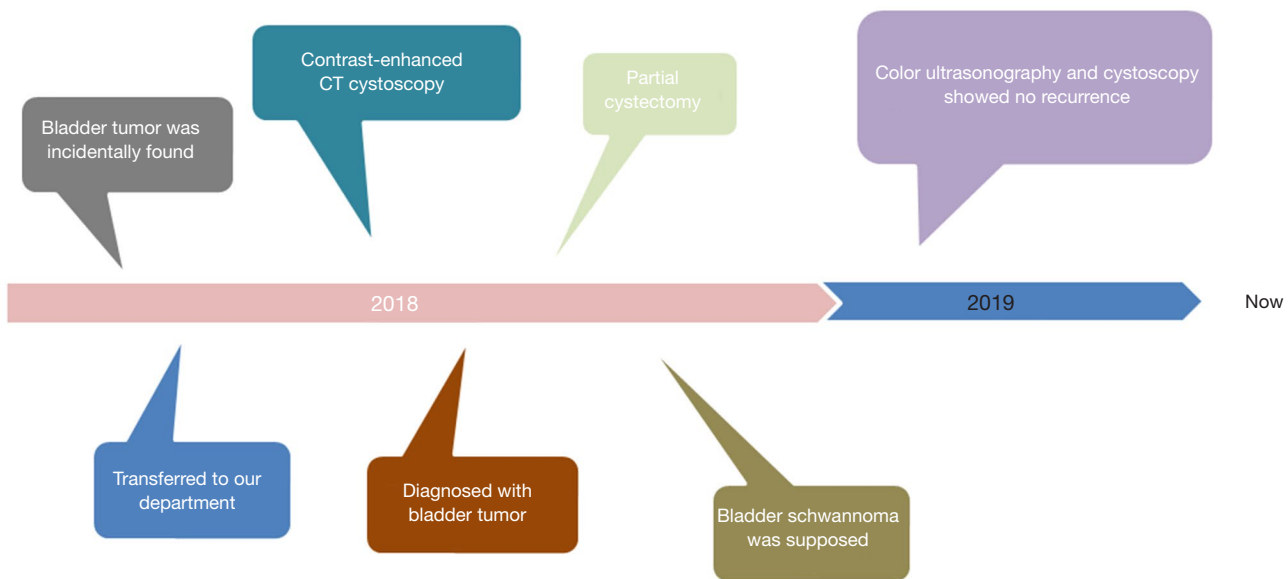


Figure 4 Timeline of interventions and outcomes.

to the above characteristics (8). On magnetic resonance imaging (MRI) scan, schwannoma presented iso-signal on T1-weighted images and slightly high signal on T2-weighted images (18). Imaging examination can not be used as a specific examination for differentiating schwannoma from other solid tumors. The cystoscopic appearance is mostly solid mass, and its surface is usually smooth, which shows significant difference from the common urothelial carcinoma. We consider that it has some significance in differential diagnosis. Pathology served as the gold standard for the diagnosis of the disease. Histologically, the iconic areas of Antoni A are also known as fasciculate area where the boundary of the cell is unclear, the nucleus is slender or oval, and the fusiform cells are closely arranged in bundles or incomplete spirals. Antoni B is also known as the reticular area, which is loosely arranged in the stroma by a small number of messy fusiform cells. Immunohistochemically, S-100 is a characteristic marker of schwannoma, which is usually positive (19).

Treatment and prognosis

Surgery is the first choice for the treatment, including transurethral resection of bladder tumor (TURBT) or partial resection. We believe that some positions that can be reached by electroendoscopy such as the bladder neck, the top or the lateral wall of the bladder are suitable for TURBT. Otherwise, partial cystectomy is recommended.

Almost all patients did not have recurrence except for the case Georgiadis G reported. The patient received TURBT and relapsed 6 months after surgery. Due to no symptoms and personal reasons, the patient was untreated, the tumor was removed again after 12 months, and the histological diagnosis confirmed residual peripheral nerve sheath tumour. Subsequent follow-up showed the tumor regrowth and conservative treatment was chosen with regular follow-up (7). In addition, radiotherapy can also be used as an attempt (10,12). It has been reported that chemotherapy is invalid to extracranial schwannoma (9). However, a patient in France received TURBT plus Botulinum toxin (botox). To our knowledge, this is the first case treated by intravesical instillation of Botox. The patient's clinical symptoms were improved. Follow-up urine cytology and pelvic MRI were normal. Moreover, this management strategy is worthy of further investigations (15). Our article comprehensively analyzed the clinical data of totally 16 patients reported in PubMed database. The follow-up period was 1.5 to 48 months.

Conclusions

The clinical feature of bladder schwannoma is not specific. In the meanwhile, imaging is difficult to distinguish from other bladder tumors. Although the shape under cystoscope has a certain hint on the disease, pathological examination is the exact standard for diagnosis. Due to the infrequent

Table 1 Reported cases of bladder schwannoma

Case	Region	Age	Sex	Presentation	Diagnostic methods	Size(cm)	Shape	Location	Management	follow up(months)
2018 Jallad <i>et al.</i> (2)	UK	25	Female	Dyspareunia	US	NA	NA	NA	TURBT	6
2012 Mosier <i>et al.</i> (6)	USA	31	Male	Pain, Hematuria	CT	1.7	Pedunculated mass	Lt lateral wall	Surgery	8
2016 Georgiadis <i>et al.</i> (7)	UK	33	Male	Haematuria	US	NA	Irregular	Bladder neck	TURBT	6
2005 Geol <i>et al.</i> (8)	Korea	35	Male	No symptom	US	3.5	Solid mass	Left lateral wall	PC	12
2020 Nasrollahi <i>et al.</i> (9)	Iran	35	Female	Frequency	US	1	Solid	Dome of bladder	TURBT	6
2016 Srinivasa <i>et al.</i> (10)	India	45	Male	Hematuria	US	1.6	Polypoid	Dome of bladder	TURBT	9
2010 Fukui <i>et al.</i> (11)	Japan	50	Female	No symptom	US	4	Solid	Anterior wall	PC	9
2014 Mazdar <i>et al.</i> (12)	Morocco	50	Female	Hematuria	US	5.8	Solid mass	Rt lateral wall	TURBT	5
2008 Gafson <i>et al.</i> (13)	UK	52	Female	Pain, vomiting, frequency	US	7	Solid mass	Anterior superior wall	Surgery	1.5
2018 Bakurov <i>et al.</i> (5)	Russia	53	Male	Hematuria, urgency	MRI	3.5	Solid mass	Bladder Neck	TURBT	12
2008 Wang <i>et al.</i> (14)	USA	56	Female	No symptom	NA	NA	Solid mass	NA	Local resection	48
2017 Zugail <i>et al.</i> (15)	France	57	Male	Urinary symptoms	Cystoscopy	NA	Flat mass	Right and left lateral walls of the bladder and dome	TURBT+BOTOX	NA
1998 Cummings <i>et al.</i> (16)	USA	58	Female	Pain, Urgency, frequency	US	4.5	Cystic lesion	Lt lateral wall	PC	36
2017 Matsumoto <i>et al.</i> (17)	Japan	68	Female	No symptom	MRI	0.8	Solid	Anterior wall	PC	6
2008 Wang <i>et al.</i> (14)	USA	69	Male	Haematuria and recurrent infection	NA	NA	Solid	NA	Local resection	48
1993 Ng <i>et al.</i> (4)	UK	88	Female	Urgency, incontinency	US	20	Solid mass	Left side	No surgery	NA
Our case	China	31	Female	No symptom	CT	3.5	Solid mass	Anterior top of the bladder	PC	29

TURBT, transurethral resection of bladder tumor; PC, partial cystectomy; US, ultrasonography; NA, not available; CT, computed tomography; MRI, magnetic resonance imaging; LT, left; RT, right.

clinical cases of bladder schwannoma, the lack of large samples and systematic research statistics, there is no unified therapeutic schedule up to the present. On account of the possibility of malignant transformation of the disease, surgical resection is the current preference. Long-term follow-up after operation is also indispensable.

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Footnote

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Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at <https://dx.doi.org/10.21037/tcr-21-200>). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient.

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