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Contents

Thrice Monthly Volume 10 Number 15 May 26, 2022

EDITORIAL

- 4713 Diet and intestinal bacterial overgrowth: Is there evidence?

Souza C, Rocha R, Cotrim HP

MINIREVIEWS

- 4717 Definition and classification of acute-on-chronic liver diseases

Zhang YY, Meng ZJ

- 4726 Management of neurosurgical patients during coronavirus disease 2019 pandemics: The Ljubljana, Slovenia experience

Velmar T, Bosnjak R

ORIGINAL ARTICLE

Clinical and Translational Research

- 4737 Glycolytic and fatty acid oxidation genes affect the treatment and prognosis of liver cancer

Zou JY, Huang YJ, He J, Tang ZX, Qin L

- 4761 Detection of a novel panel of 24 genes with high frequencies of mutation in gastric cancer based on next-generation sequencing

Zeng HH, Yang Z, Qiu YB, Bashir S, Li Y, Xu M

Case Control Study

- 4776 Outcomes of cervical degenerative disc disease treated by anterior cervical discectomy and fusion with self-locking fusion cage

Zhang B, Jiang YZ, Song QP, An Y

- 4785 Impact of COVID-19 pandemic on clinicopathological features of transplant recipients with hepatocellular carcinoma: A case-control study

Akbulut S, Sahin TT, Ince V, Yilmaz S

Retrospective Study

- 4799 Risk factors and optimal predictive scoring system of mortality for children with acute paraquat poisoning

Song Y, Wang H, Tao YH

- 4810 Application effect of thoracoscopic tricuspid valvuloplasty in geriatric patients with tricuspid valve disease

Jiang W, Long XM, Wei KQ, Li SC, Zhang Z, He BF, Li H

- 4818 Endoscopic ultrasonography in the evaluation of condition and prognosis of ulcerative colitis

Jin RF, Chen YM, Chen RP, Ye HJ

- 4827** Dynamic interaction nursing intervention on functional rehabilitation and self-care ability of patients after aneurysm surgery

Xie YE, Huang WC, Li YP, Deng JH, Huang JT

Clinical Trials Study

- 4836** Validations of new cut-offs for surgical drains management and use of computerized tomography scan after pancreatoduodenectomy: The DALCUT trial

Caputo D, Coppola A, La Vaccara V, Passa R, Carbone L, Ciccozzi M, Angeletti S, Coppola R

Observational Study

- 4843** Psychosocial adaptation and influencing factors among patients with chemotherapy-induced peripheral neuropathy

Zhou X, Wang DY, Ding CY, Liu H, Sun ZQ

META-ANALYSIS

- 4856** Outcome of the efficacy of Chinese herbal medicine for functional constipation: A systematic review and meta-analysis

Lyu Z, Fan Y, Bai Y, Liu T, Zhong LL, Liang HF

CASE REPORT

- 4878** Familial gastrointestinal stromal tumors with *KIT* germline mutation in a Chinese family: A case report

Yuan W, Huang W, Ren L, Xu C, Luan LJ, Huang J, Xue AW, Fang Y, Gao XD, Shen KT, Lv JH, Hou YY

- 4886** Nonfunctional pancreatic neuroendocrine tumours misdiagnosed as autoimmune pancreatitis: A case report and review of literature

Lin ZQ, Li X, Yang Y, Wang Y, Zhang XY, Zhang XX, Guo J

- 4895** Sudden deafness as a prodrome of cerebellar artery infarction: Three case reports

Li BL, Xu JY, Lin S

- 4904** Importance of abdominal X-ray to confirm the position of levonorgestrel-releasing intrauterine system: A case report

Maebayashi A, Kato K, Hayashi N, Nagaishi M, Kawana K

- 4911** Bedside ultrasonic localization of the nasogastric tube in a patient with severe COVID-19: A case report

Zhu XJ, Liu SX, Li QT, Jiang YJ

- 4917** Paradoxical herniation after decompressive craniectomy provoked by mannitol: A case report

Du C, Tang HJ, Fan SM

- 4923** Targeted next-generation sequencing identifies a novel nonsense mutation in ANK1 for hereditary spherocytosis: A case report

Fu P, Jiao YY, Chen K, Shao JB, Liao XL, Yang JW, Jiang SY

- 4929** Nonfunctional bladder paraganglioma misdiagnosed as hemangioma: A case report

Chen J, Yang HF

- 4935** Special type of Werneck syndrome in midbrain infarction: Four case reports
Yang YZ, Hu WX, Zhai HJ
- 4942** Primary extraskeletal Ewing's sarcoma of the lumbar nerve root: A case report
Lei LH, Li F, Wu T
- 4949** Yellow nail syndrome accompanied by minimal-change nephrotic syndrome: A case report
Zhang YN, Wang MH, Yu WC, Cheng W, Cong JP, Huang XP, Wang FF
- 4957** Total femur replacement with 18 years of follow-up: A case report
Yang YH, Chen JX, Chen QY, Wang Y, Zhou YB, Wang HW, Yuan T, Sun HP, Xie L, Yao ZH, Yang ZZ
- 4964** Male metaplastic breast cancer with poor prognosis: A case report
Kim HY, Lee S, Kim DI, Jung CS, Kim JY, Nam KJ, Choo KS, Jung YJ
- 4971** CD8-positive indolent T-Cell lymphoproliferative disorder of the gastrointestinal tract: A case report and review of literature
Weng CY, Ye C, Fan YH, Lv B, Zhang CL, Li M
- 4985** Bone flare after initiation of novel hormonal therapy in patients with metastatic hormone-sensitive prostate cancer: A case report
Li KH, Du YC, Yang DY, Yu XY, Zhang XP, Li YX, Qiao L
- 4991** Postoperative infection of the skull base surgical site due to suppurative parotitis: A case report
Zhao Y, Zhao Y, Zhang LQ, Feng GD
- 4998** Blunt aortic injury-traumatic aortic isthmus pseudoaneurysm with right iliac artery dissection aneurysm: A case report
Fang XX, Wu XH, Chen XF
- 5005** Extensive complex thoracoabdominal aortic aneurysm salvaged by surgical graft providing landing zone for endovascular graft: A case report
Jang AY, Oh PC, Kang JM, Park CH, Kang WC
- 5012** Gastric heterotopia of colon found cancer workup in liver abscess: A case report
Park JG, Suh JI, Kim YU
- 5018** Clinical manifestations and gene analysis of Hutchinson-Gilford progeria syndrome: A case report
Zhang SL, Lin SZ, Zhou YQ, Wang WQ, Li JY, Wang C, Pang QM
- 5025** Neurocutaneous melanosis with an intracranial cystic-solid meningeal melanoma in an adult: A case report and review of literature
Liu BC, Wang YB, Liu Z, Jiao Y, Zhang XF
- 5036** Metastasis of liver cancer to the thyroid after surgery: A case report
Zhong HC, Sun ZW, Cao GH, Zhao W, Ma K, Zhang BY, Feng YJ

- 5042** Spontaneous liver rupture following SARS-CoV-2 infection in late pregnancy: A case report
Ambrož R, Stašek M, Molnár J, Špička P, Klos D, Hambálek J, Skanderová D
- 5051** Carotid blowout syndrome caused by chronic infection: A case report
Xie TH, Zhao WJ, Li XL, Hou Y, Wang X, Zhang J, An XH, Liu LT
- 5057** Is repeat wide excision plus radiotherapy of localized rectal melanoma another choice before abdominoperineal resection? A case report
Chiu HT, Pu TW, Yen H, Liu T, Wen CC
- 5064** Metaplastic breast cancer with chondrosarcomatous differentiation combined with concurrent bilateral breast cancer: A case report
Yang SY, Li Y, Nie JY, Yang ST, Yang XJ, Wang MH, Zhang J
- 5072** Rare solitary splenic metastasis from a thymic carcinoma detected on fluorodeoxyglucose-positron emission tomography: A case report
Tsai YH, Lin KH, Huang TW
- 5077** Type A aortic dissection following heart transplantation: A case report
Zeng Z, Yang LJ, Zhang C, Xu F
- 5082** Catheter-related infections caused by *Mycobacterium abscessus* in a patient with motor neurone disease: A case report
Pan SF, Zhang YY, Wang XZ, Sun JJ, Song SL, Tang YR, Wang JL
- 5088** Clear aligner treatment for a four-year-old patient with anterior cross-bite and facial asymmetry: A case report
Zou YR, Gan ZQ, Zhao LX
- 5097** Knot impingement after arthroscopic rotator cuff repair mimicking infection: A case report
Kim DH, Jeon JH, Choi BC, Cho CH
- 5103** Solitary primary pulmonary synovial sarcoma: A case report
He WW, Huang ZX, Wang WJ, Li YL, Xia QY, Qiu YB, Shi Y, Sun HM
- 5111** Anesthetic management for intraoperative acute pulmonary embolism during inferior vena cava tumor thrombus surgery: A case report
Hsu PY, Wu EB
- 5119** Delayed diagnosis of arytenoid cartilage dislocation after tracheal intubation in the intensive care unit: A case report
Yan WQ, Li C, Chen Z

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Nonfunctional bladder paraganglioma misdiagnosed as hemangioma: A case report

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Abstract

BACKGROUND

Paraganglioma of the urinary bladder (PUB) is a nonepithelial tumor of the bladder that is also known as bladder pheochromocytoma. The incidence of nonfunctional paraganglioma of the urinary bladder is rare in clinical practice.

CASE SUMMARY

A case of nonfunctional paraganglioma of the urinary bladder diagnosed and treated in our hospital is reported herein. A 2.5 cm × 2.1 cm moderate-echo mass protruding into the right anterior wall of the bladder was incidentally found during transvaginal color Doppler ultrasound examination. Based on a contrast-enhanced computed tomography scan of the bladder, the right anterior wall of the bladder was considered to present a hemangioma. The patient underwent laparoscopic partial resection of the bladder wall in our hospital, and pathological results showed paraganglioma of the bladder. At present, we have followed up with the patient for half a year, and there has been no recurrence of the tumor.

CONCLUSION

The imaging patterns of PUB and bladder hemangioma are similar. In clinical practice, the possibility of PUB should be considered for a single wide-based isoechoic mass in the bladder, especially functional PUB may cause some adverse effects during surgery.

Key Words: Bladder tumor; Paraganglioma; Diagnosis and treatment methods; Case report

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Core Tip: The imaging patterns of paraganglioma of the urinary bladder (PUB) and bladder hemangioma are similar. In clinical practice, the possibility of PUB should be considered for a single wide-based isoechoic mass in the bladder, especially functional PUB may cause some adverse effects during surgery. This case presents a case of nonfunctional PUB misdiagnosed in our medical center. The pathogenesis, clinical manifestations and imaging findings of PUB are also described in order to improve everyone's understanding of PUB.

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INTRODUCTION

Paraganglioma of the urinary bladder (PUB) is a rare bladder neoplasm subtype, accounting for approximately 0.06% of bladder tumors. There are significant differences in symptoms between individuals, which makes this tumor extremely easy to misdiagnose or miss. A case of nonfunctional PUB diagnosed and treated in our hospital is reported herein.

CASE PRESENTATION

Chief complaints

A 22-year-old female patient was hospitalized on February 14, 2021, because "the bladder was found to feel occupied for 5 d."

History of present illness

The patient underwent transvaginal ultrasonography for routine premarital examination 5 d prior. During the examination, a 2.5 cm × 2.1 cm medium-echo mass protruding into the right anterior wall of the bladder was found. The shape was regular. The boundary was clear, and the wide base was connected to the bladder wall. Color Doppler flow imaging detected rich strip blood flow signals, with an RI of 0.66 (Figure 1).

History of past illness

The patient had a free previous medical history.

Personal and family history

The patient was previously healthy and her family had no relevant medical history.

Physical examination

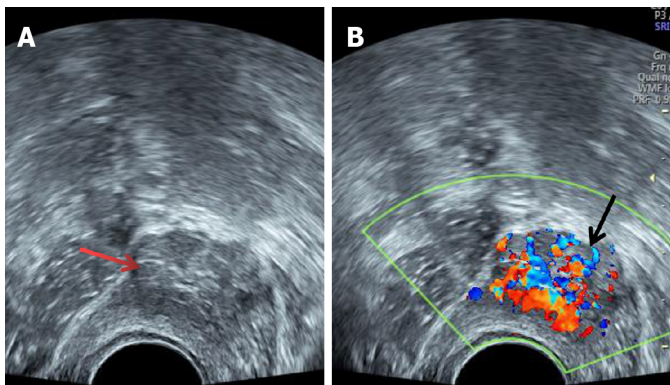
The patient had no frequent urination, urgency dysuria, or gross hematuria. There were no symptoms, such as hypertension, palpitations or cold sweats, during urination. Upon the physical examination, the patient's heart rate was 75 bpm, and her blood pressure was 110/68 mmHg. There was no eminence, tenderness or percussion pain in the bladder area.

Laboratory examinations

Blood analysis, blood biochemistry and urinalysis revealed no significant abnormalities. Electrocardiogram and abdominal ultrasound (except bladder) also showed no abnormality.

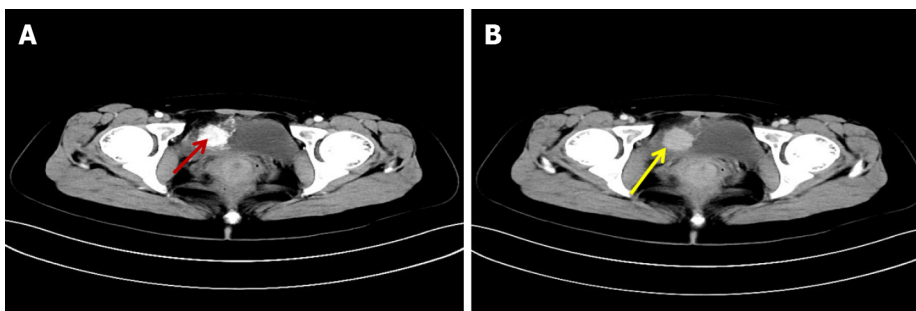
Imaging examinations

An enhanced computed tomography (CT) scan of the bladder was performed after admission: the right front of the bladder was occupied, and the boundary with the bladder wall was not clear. The bladder was under pressure, and the size was approximately 2.5 cm × 2.4 cm. In the arterial phase of the enhanced scan, obvious enhancement could be seen as well as multiple tortuous vascular shadows. The lesion was considered a hemangioma (Figure 2).



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Figure 1 Transvaginal ultrasound showed a 2.5 cm × 2.1 cm medium-echo mass protruding from the right anterior wall of the bladder (A, indicated by the red arrow); a regular shape, a clear boundary, a wide base, and rich strip blood flow signals were detected (B, indicated by the black arrow).



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Figure 2 Enhanced computed tomography scan of the bladder showed a circular space-occupying lesion on the right anterior wall of the bladder, with obvious enhancement in the arterial phase (A, indicated by the red arrow) and weakening of the enhancement in the delayed phase (B, indicated by the yellow arrow).

FINAL DIAGNOSIS

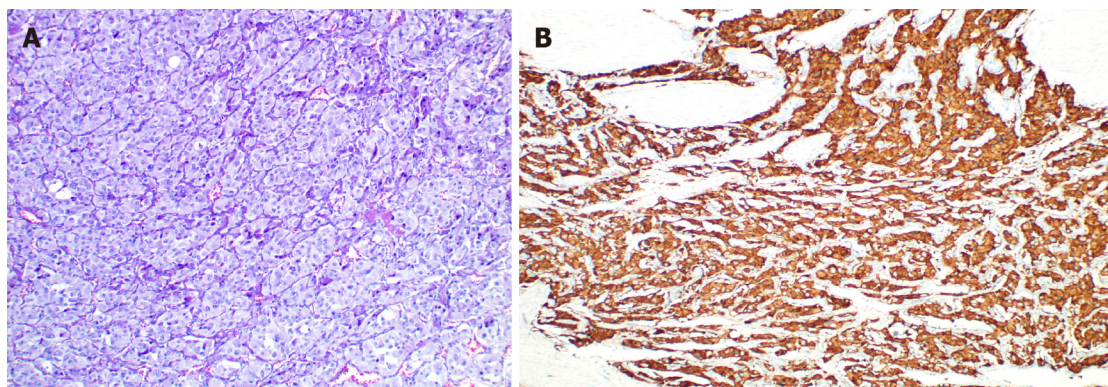
Nonfunctional paraganglioma of the urinary bladder.

TREATMENT

When perfecting the relevant examinations and excluding surgical contraindications, the patient underwent laparoscopic partial cystectomy under general anesthesia. During the operation, a space-occupying lesion on the right side of the bladder outside the peritoneum was found, with a smooth surface. The space invaded the bladder wall, and its border was unclear. The bladder wall connected to the new mass was removed under direct laparoscopic vision. The bladder incision was then sutured. After observing that there was no obvious bleeding from the incision, an abdominal drainage tube was placed, a 16 F three-chamber balloon tube was placed in the bladder, and 10 mL of water was injected. The procedure went smoothly. The patient had no obvious discomfort during the operation, with normal vital signs.

OUTCOME AND FOLLOW-UP

The catheter was removed 14 d after the operation. Postoperative pathological findings indicated paraganglioma of the bladder. The cut surface of the lump was grayish-white and gray-brown, and the texture is slightly hard. Microscopically, the tumor cells were polygonal, with large nuclei, visible nucleoli, a nest-like arrangement, and abundant blood vessels. The immunohistochemical staining results were as follows: Syn (+), CgA (+), CD56 (+), S-100 (supporting cells+), CK (-), GATA3 (-), and Ki-67 (+, < 2%) (Figure 3). The patient's 24-h urinary CA, plasma-free MNs and 24-h urinary vanillyl-



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Figure 3 Pathological and immunological features of a paraganglioma of the urinary bladder. A: Micrograph of conventional HE staining (× 200) showing chief cells in a nested growth pattern; B: The image shows positive expression of CgA in immunohistochemistry.

mandelic acid (VMA) improved, and the results were normal. During the 6 mo of follow-up, there was no recurrence or metastasis of the tumor, and the patient did not complain of discomfort.

DISCUSSION

PUB is a neuroendocrine tumor that originates from the chromaffin tissue of the sympathetic nerve in the bladder wall[1]. PUB mainly occurs in adults[2] and is mostly located on the sidewall of the bladder; the trigone has the least common incidence[3], which may be related to the distribution of chromaffin tissue on the bladder wall. PUB can be divided into functional and nonfunctional types according to whether the tumor secretes catecholamine hormones and the release activity of hormones[4,5]. Functional PUB can manifest as a clinical type or a resting type. Hypertension, dizziness, palpitations and even syncope during urination are the specific manifestations of clinical PUB. Compared with resting PUB, the symptoms of the clinical type are more subtle. Only touching or cutting the tumor during examination or surgery can cause a large amount of catecholamines to enter the bloodstream, which causes the patient to develop symptoms. Nonfunctional PUB primarily manifests as painless hematuria and urinary tract obstruction or is found during physical examination. In the present case, the patient did not have any clinical symptoms. Indeed, the mass was only discovered by chance during a gynecological physical examination. There was no clinical manifestation of large amounts of catecholamines in the blood when touching the tumor during the operation or after the operation. In addition, the catecholamine hormone levels in the blood and urine of the patient measured after the operation were within the normal ranges. Therefore, it was diagnosed as nonfunctional PUB.

Qualitative diagnosis of paraganglioma mainly depends on the detection of blood and urine catecholamines and 24-h urine VMA. Location diagnosis relies on imaging [Ultrasound, CT and magnetic resonance imaging (MRI)] and cystoscopy. Ultrasound is the most routinely used method for the urinary system. PUB shows a hypoechoic or isoechoic mass with a wide base on two-dimensional ultrasound. Color Doppler ultrasound can elicit abundant blood flow signals in PUB[5,6]. PUB on plain CT scan imaging is a single round or round-like soft tissue mass on the lateral wall of the bladder. It may be accompanied by necrotic cystic degeneration and arc-shaped calcification. Based on enhanced CT scans, PUB is significantly enhanced in the arterial phase, with weakened enhancement in the delayed phase, which is its typical feature; arterial shadows are occasionally visible[6]. This feature is of great significance for preoperative localization and qualitative diagnosis. In contrast, most bladder urothelial carcinomas show mild to moderate enhancement on enhanced CT scans[7]. PUB display an equal or slightly high signal on T1WI in MRI and a high "salt and pepper" signal on T2WI; on DWI, it shows a high signal. PUB shows significant enhancement after intravenous injection of Gd-DTPA[6,8]. Radionuclide-labeled metaiodobenzyl guanidine (MIBG) imaging can combine the functional characteristics of pheochromocytoma with morphological changes with high accuracy and specificity[9]. PUB showed hypermetabolic lesions on ¹³¹I MIBG SPECT/CT. Nuclear medicine examination can accurately identify the location and size of lesions and can perform staging determination and treatment[10] (Table 1).

In our case, the first lesion was found on vaginal ultrasound. Because the probe was placed in the vaginal vault, we could comprehensively observe the bladder and urethra during the vaginal ultrasound scan, and the results were clearer than those of transabdominal ultrasound. The transvaginal ultrasound findings were consistent with those previously reported based on transabdominal ultrasound[5,6,11]. A wide-base isoechoic round mass with abundant blood flow signals was detected. During enhanced CT, we also noticed a single lesion on the bladder wall. After enhancement, the signal

Table 1 Clinical manifestations and related examination results of paraganglioma of the urinary bladder

Clinical manifestations	Most nonfunctional PUB have no obvious symptoms; functional PUB is often accompanied by posturination syndrome such as hypertension and palpitations
Radiology	
Ultrasound	Hypoechoic or isoechoic mass in two-dimensional, hypervascular mass by color Doppler ultrasound.
CT	Unenhanced scan showed a round mass that was convex to the cavity, with obvious enhancement in the arterial phase during enhancement and weak enhancement in the delayed phase
MRI	All tumors exhibited slight hyperintensity on T1WI and hyperintensity with "salt and pepper" appearance on T2WI, with a strong high signal on DWI
Nuclear medicine	¹³¹ I MIBG SPECT/CT combines the functional and morphological changes of PUB, with high sensitivity and specificity
Laboratory tests	Some may have increased 24-h urinary catecholamines and urinary VMA
Cystoscopy	Occurs in the lateral wall, posterior wall, anterior wall and the top of the bladder; there is no obvious abnormality in early examination, but in the late stage, it is often accompanied by hyperemia, calcification or necrosis of the bladder mucosa

PUB: Paraganglioma of the urinary bladder; VMA: Vanillylmandelic acid; CT: Computed tomography; MRI: Magnetic resonance imaging; MIBG: Metaiodobenzyl guanidine.

was obviously strengthened and then slowly subsided in the delay period; thus, it was considered to be a hemangioma. However, most patients with bladder hemangioma have recurrent painless hematuria [12]. In addition, sessile, blue, raised masses were found by cystoscopy [13]. Although the two can be indistinguishable on imaging, they can be distinguished by combining clinical symptoms and other auxiliary examinations.

The bladder is the most conventional site of urogenital paragangliomas (79.2%). Most PUBs are histologically benign, and only approximately 10% of PUB cases are malignant [14]. At present, there is no unified standard for the treatment of paraganglioma of the urinary bladder, but surgery is still the main method. Surgical methods include transurethral resection of bladder tumor, partial cystectomy and radical cystectomy. The specific choice of surgical method should be based on the actual situation of the patient and the technical strength of the surgeon to reduce risk and ensure safety as much as possible. In this case, laparoscopic partial cystectomy was performed because the mass did not protrude into the bladder cavity and the broad base was connected to the bladder wall.

In summary, PUB is very uncommon. It is misdiagnosed easily and missed clinically. For the bladder appears a single round, well-circumscribed and wide base of the mass, the possibility of PUB should be considered before surgery. Surgeons should be fully prepared. Patients should be followed up for a long time after surgery. There are currently no histological, genetic or molecular markers that can distinguish benign and malignant paragangliomas preoperatively. The diagnosis of a malignant tumor relies only on the presence of metastasis.

CONCLUSION

The imaging patterns of PUB and bladder hemangioma are similar. In clinical practice, the possibility of PUB should be considered for a single wide-based isoechoic mass in the bladder, especially functional PUB may cause some adverse effects during surgery. Surgeons should be fully prepared. Patients should be followed up for a long time after surgery so that recurrence of the disease can be detected early.

FOOTNOTES

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