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Intraoperative decision to change the course of management based on an ultrasonographic image of urinary bladder paraganglioma – a case study

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Abstract

This case study concerns a 69-year-old woman qualified for transurethral resection of a primary tumor in a urinary bladder. Since a cystoscopic image did not correspond with typical urothelial carcinoma, an intraoperative ultrasonographic transrectal imaging was carried out, on the basis of which it was believed to be urinary bladder paraganglioma. Endoscopic treatment was abandoned. The patient was directed for further examination and next, on suspicion of extra-bladder pheochromocytoma, scheduled for open surgery. Due to the location and range of the tumor, and systemic conditions, a radical cystectomy and Bricker's supravescical ureteroileal conduit were carried out. A postoperative histopathological examination confirmed the pheochromocytoma character of the urinary bladder tumor.

Case report

A 69-year-old patient was admitted to a urinary ward due to a primary tumor of the urinary bladder. An exophytic tumor was found in a trans-abdominal ultrasonographic examination conducted after an episode of asymptomatic hematuria. Based on the ultrasonographic examination, the patient was qualified for a transurethral electro-resection of the tumor. During the surgery, a tumor ca. 25 mm in diameter, with a smooth surface and a rich network of curved submucosal vessels was found on a posterior bladder wall (Fig. 1). Since a cystoscopic image did not correspond with typical urothelial lesion, an intraoperative ultrasonographic transvaginal examination was carried out (Fig. 2, Fig. 3, Fig. 4).

In the ultrasound examination, a tumor 25 mm in diameter infiltrating the urinary bladder wall toward the vagina anterior wall was imaged. In color Doppler, a signal of very rich vascular flow was registered, which is untypical for urothelial tumors (Fig. 5, Fig. 6).

As an incomplete transurethral resection and a risk of fistula between the urinary bladder and vagina development were anticipated, transurethral resection was abandoned. Due to very rich vascularization of the lesion, intensive bleeding from the tumor was anticipated, and so a biopsy with a resectoscope loop was not carried out. Also, transvaginal core needle biopsy was not conducted, since the specimens would be of low quality due to delicate and vascular structure of the lesion.

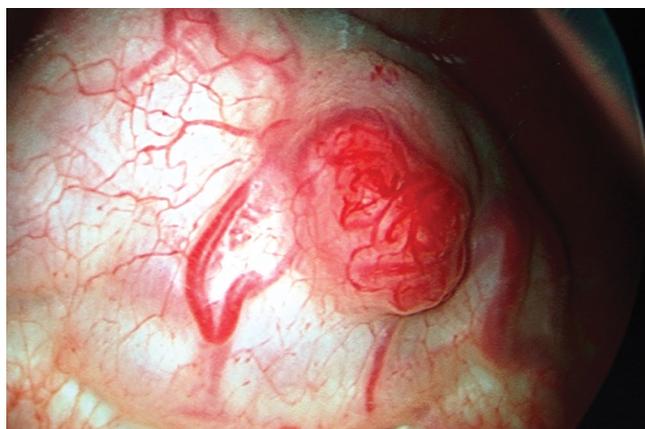


Fig. 1. Cystoscopic image of a urinary bladder tumor. A rich submucosal vascular network is conspicuously displayed; mucosa adjacent to tumor is unchanged



Fig. 2. Urinary bladder image in trans-abdominal examination in transverse plain. Tumor infiltrating through a bladder wall and adjacent to a cervix. Examination conducted with ProFocus Ultraview, BK Medical, 8820e probe, 5 MHz

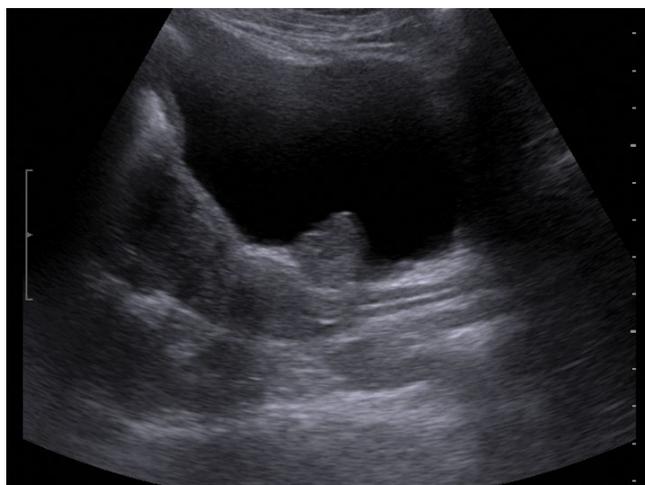


Fig. 3. Urinary bladder image in transabdominal examination in sagittal plain. Tumor infiltrating through a bladder wall and adjacent to cervix. Examination conducted with ProFocus Ultraview, BK Medical, 8820e, 5 MHz probe

Additionally, resection and biopsy were abandoned due to the suspicion that the tumor was pheochromocytoma, and surgical intervention without proper preoperative evaluation and preparation of the patient might result in hypertension crisis. Furthermore, due to the suspicion of the tumor's extra-bladder infiltration, open surgery should be considered, to which the patient did not consent.

After the surgery, the patient was informed about the reasons for transurethral resection abandoning and the need for further examination (Fig. 7, Fig. 8, Fig. 9). A CT examination of the abdomen and the pelvis was carried out (Fig. 10, Fig. 11), as well as a MR examination of the pelvis (Fig. 12, Fig. 13). The patient was referred for an endocrinological consultation.

The patient was scheduled for open surgery – radical hysterectomy and resection of the adjacent part of the urinary bladder with the tumor.

Eventually, radical cystectomy and uterus resection with adnexa were performed, and a ureteroileal conduit created. The postoperative period was uneventful.

In the post-operative histopathological examination, urinary bladder paraganglioma was found. The following immunohistochemical reactions were investigated: chromogranin+, synaptophysin+, vimentin+, S100+, NSE+, CK–, CK7–, CK20–, CK5/6–, CD10–, HMW CK–. The tumor occupied almost the entire thickness of the urinary bladder muscle. Spreading of the tumor onto bladder-adjacent tissues was not reported. Necrosis or mitoses centers were not observed in the tumor's profile.

Discussion

Paragangliomas or extra-adrenal pheochromocytoma make up ca. 15% of all pheochromocytoma. Urinary bladder paragangliomas make up ca. 6% of all paraganglioma and 0,06% of all urinary bladder tumors⁽¹⁾.

The patient was planned (scheduled) for a transurethral electro-resection of a bladder tumor found during a trans-abdominal ultrasound examination of the urinary bladder. In a trans-abdominal examination carried out in a different center, an exophytic sessile lesion without surface calcifications was found. Such image may correspond with uroepithelial cancer. In the case of urothelial carcinomas, episodes of intensive hematuria are frequent. Additionally, for over 40 years the patient had smoked at least 20 cigarettes a day. Therefore, she was at risk of urothelial tumor development. The patient did not report any episodes of high blood pressure, heartbeat acceleration or malaise, these may be linked with the presence of urinary bladder paraganglioma⁽²⁾, due to excessive bladder filling during miction or after it. There were episodes of high blood pressure up to 180/100 mmHg which were, however, associated with irregular intake of antihypertensive drugs.

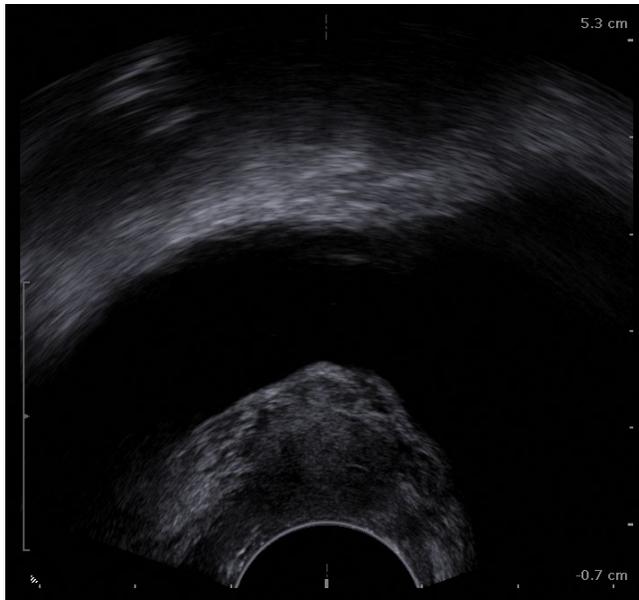


Fig. 4. Urinary bladder image in transvaginal examination in transverse plain. Tumor seems to invade a vaginal wall. Examination conducted with ProFocus Ultraview, BK Medical, a 8818, 12 MHz probe

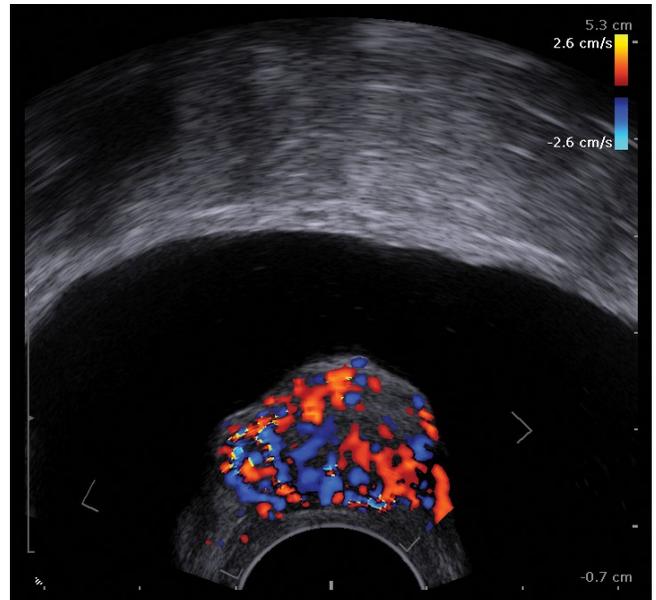


Fig. 5. Urinary bladder image in transvaginal examination in transverse plain in color Doppler. A rich submucosal vascular network, which supplies the tumor, is conspicuously displayed. Examination conducted with ProFocus Ultraview, BK Medical, 8818, 12 MHz probe

In a cystoscopic image, posteriorly to bladder trigone, a tumor of 20–25 mm in diameter was found. It was covered with normal mucosa, yet with a well-developed network of submucosal vessels. In the differentiation, it was necessary to take into consideration tumors invading the urinary bladder from the side of the reproductive organ, metastases from other organs, as well as muscle or connective tissue-based cancers

including vascular tumors and tumors originating from ganglions⁽³⁾.

According to an intraoperative ultrasound examination, there was no need for a detailed assessment of the change's size and its relation to the neighboring structures in terms of the possibility of radical transurethral resection. Although the results of a trans-abdominal examination suggested that the tumor spread through the whole thickness of the bladder's wall, it was only the transvaginal ultrasonography that revealed the tumor not only had invaded the bladder's wall but also revealed the

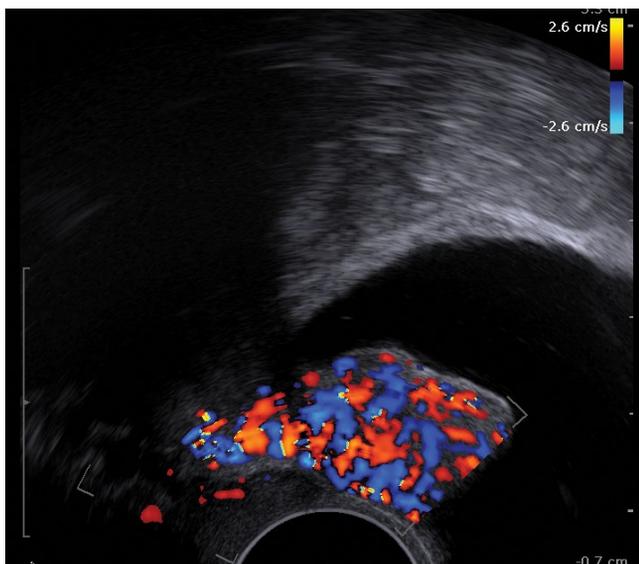


Fig. 6. Urinary bladder image in trans-vaginal examination in sagittal plain in color Doppler. A rich submucosal vascular network, which supplies tumor from the side of cervix, is conspicuously displayed. Examination conducted with ProFocus Ultraview, BK Medical, a 8818, 12 MHz probe

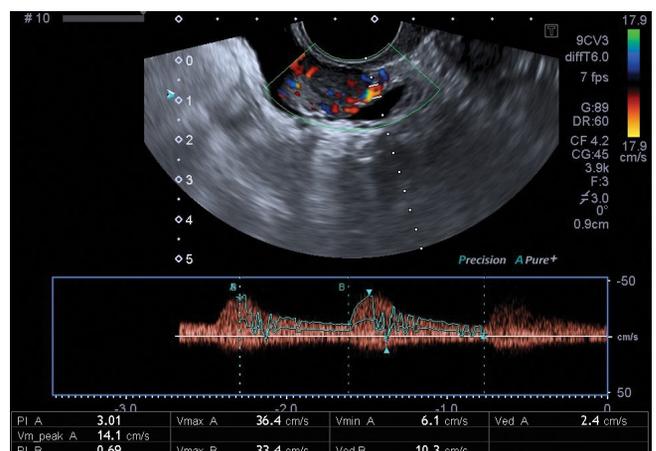


Fig. 7. Urinary bladder image in transvaginal examination in sagittal plain in color Doppler with a blood flow spectrum in a blood vessel supplying tumor (carried out a few days after intraoperative examination). Examination conducted with Toshiba Aplio 500, an endovaginal 9 MHz probe

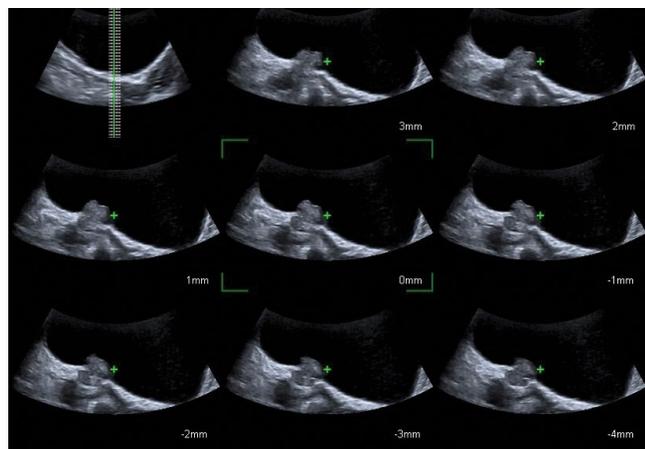


Fig. 8. Urinary bladder scans in trans-abdominal examination with a volumetric head allow for precise assessment of tumor's extent in relation to a uterus. Examination conducted with Toshiba Aplio 500, an endovaginal 9CV3 probe

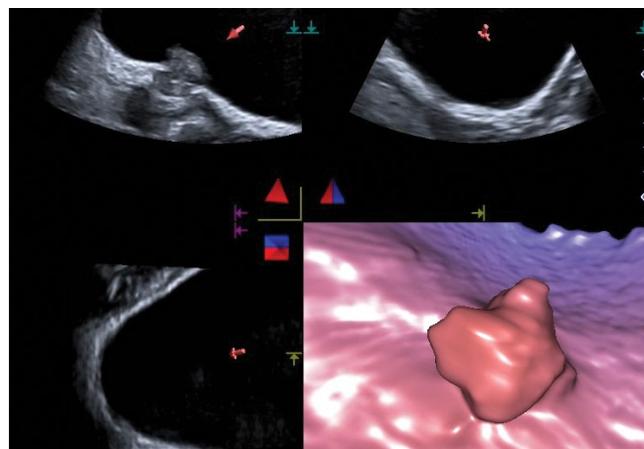


Fig. 9. Three-dimensional reconstruction of a urinary bladder wall with tumor carried out with volumetric scanning probe. Ultra-sonographic visualization of urinary bladder wall – sonocystoscopy accurately corresponds with cystoscopy. Examination conducted with Toshiba Aplio 500, an endovaginal 9CV3 probe

risk of it invading the vagina's and cervix' anterior walls. In such a situation, an attempt of transurethral resection would result either in the tumor's non-radical resection or development of a fistula between the urinary bladder and the vagina. According to the Doppler examination findings, the tumor had a very dense vascular network and very intensive blood flow, which was inadequate to the change's size. It might suggest angioma or urinary bladder paraganglioma. Based on an ultrasound examination, it was impossible to decide whether the change's rich vascularization was of an angioma or urinary bladder paraganglioma type. In the case of minor angiomas, endoscopic biopsy, resection and fulguration are safe and do not lead to relapses⁽⁴⁾, yet in the case of phaeochromocytoma, a similar procedure may result in a life-threatening hypertensive crisis. An attempt at a paraganglioma transurethral partial resection, performed in order to

collect specimens for a histopathological examination, would have resulted in intensive intraoperative bleeding and thus, force deep coagulation of the bladder's wall, which had been witnessed by the surgeon in the past⁽⁵⁾. On the other hand, transvaginal core needle biopsy from a transparent vascular tumor would not have guaranteed obtaining specimens of satisfactory quality for a histopathological assessment.

The patient was immediately referred for computer tomography of the abdomen and pelvis, and a magnetic resonance of the pelvis, the results of which were concurrent with a transvaginal ultrasound assessment. The patient was directed to an endocrinological ward for the assessment of hormonal activity, a scintigraphy examination of an adrenergic system with metaiodobenzylguanidine marked with radioactive iodine (MIBG-J131),



Fig. 10. CT examination with intravenous injection of a contrast medium, arterial phase, axial section. On the right in the wall of urinary bladder, a smoothly outlined structure with intensive enhancement is visible, which corresponds with phaeochromocytoma



Fig. 11. CT examination with intravenous injection of a contrast medium, delayed phase after 15 minutes, axial section. On the right in the wall of urinary bladder, a filling defect is visible, which corresponds with phaeochromocytoma

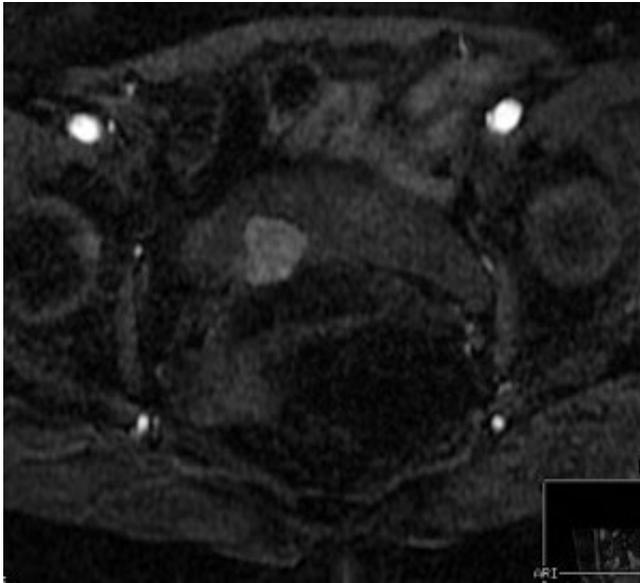


Fig. 12. Phaeochromocytoma in magnetic resonance examination in axial section. In T1-dependant sequence of gradient echo with adipose tissue saturation and intravenous application of paramagnetic contrast medium, presence of phaeochromocytoma with intensive enhancement was confirmed

and implementation of proper hypertension treatment⁽⁶⁾ as a part of preparation for open surgery. In a specialist center for oncological urology, radical cystectomy, uterus resection with adnexa, and ureteroileal conduit modo Bricker were carried out. Postoperative period was uneventful. Scope of the surgery resulted from the suspicion of the invasion of the structures adjacent to

the bladder and proximity of distal parts of ureters and ureteral orifices. In the case of paragangliomas located far from a urinary bladder triangle, partial resection of a urinary bladder should be considered⁽⁷⁾. In the case of pheochromocytoma located on a side wall nearby a ureteral orifice or distal part of the ureter, resection of the lesion may compel ureteral reimplantation into a normal bladder wall. What also should be taken into consideration in the case of the resection of paragangliomas located near an intraureteric ridge or within a bladder trigone, is the reimplantation of ureters whose orifices or distal parts could be damaged during surgery or become occluded as a result of cicatrizing in the process of post-operative recovery. If the operation on a bladder leads to a significant decrease in its volume, it may be completed with an appropriately prepared vascularized fragment of an intestinal loop. When resection of the whole bladder is necessary, creation of an alternate intestinal urinary tract should be considered. In this case, the patient's cognitive limitations were contraindications to this solution⁽⁸⁾.

In a post-surgical histopathological examination, paraganglioma was found, which occupied almost the entire thickness of the urinary bladder muscle layer. Metastasization of the tumor onto bladder-adjacent tissues was not reported. Necrosis or mitoses centers were not observed in the tumor's profile.

This description of paraganglioma is the first in which cystoscopic appearance of the tumor and an ultrasound examination findings resulted in a suspicion of paraganglioma and change to proper course of management.

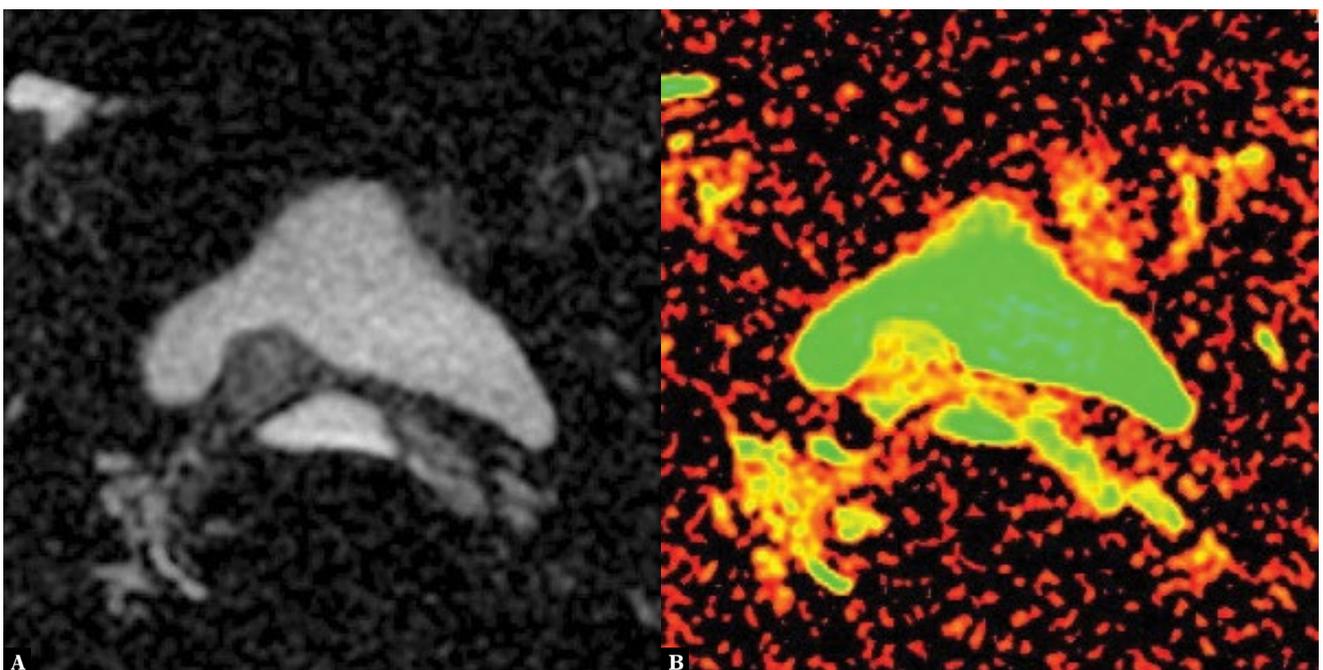


Fig. 13. Phaeochromocytoma in diffusion imaging (two b values:50, 600), in correlation with apparent diffusion coefficient map (ADC); ratio calculated automatically with diagnostic station software. Axial scans image pathological tissue with moderate traits of water diffusion restriction

Conflict of interest

Authors do not declare any financial or personal relations with other people or organizations which may negatively influence the content of this publication or claim the right to this publication.

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