Case Reports in Oncology

Case Rep Oncol 2024;17:484–489
DOI: 10.1159/000538073
Received: December 21, 2023
Accepted: January 25, 2024
Published online: March 25, 2024

Case Report

Bladder Paraganglioma Presenting as Post-Micturition Palpitations: A Case Report

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Keywords
Urinary bladder tumor · Pheochromocytoma · Neuroendocrine tumor · Paraganglioma · Case report

Abstract
Introduction: Paraganglioma of the urinary bladder (PUB) is an extremely rare extra-adrenal catecholamine-secreting neuroendocrine tumor, accounting for only 0.05% of all bladder tumors and 1% of all pheochromocytomas. The clinical presentation of PUB can be diverse and challenging to diagnose. ***Case Presentation:*** This case report presents a 37-year-old man with post-micturition palpitation, headache, and sweating, with no history of hematuria or other irritative urinary symptoms. Ultrasound and contrast-enhanced computed tomography revealed a suspicious mass in the right posterolateral wall of the urinary bladder. Despite normal functional hormonal tests, the diagnosis of PUB was confirmed after surgical enucleation and histopathological examination. ***Conclusion:*** This report emphasizes the importance of considering PUB as a differential diagnosis in patients with post-micturition symptoms and paroxysmal hypertension, as well as the need for a multidisciplinary approach in the evaluation and management of such rare and complex cases. Early recognition and surgical intervention are crucial for optimal management and favorable clinical outcomes.

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Introduction

Pheochromocytoma is a rare catecholamine-secreting neuroendocrine tumor predominantly arising from chromaffin cells in the adrenal medulla. However, approximately 10% of these tumors can arise from extra-adrenal locations, known as paragangliomas [1, 2]. Paraganglioma of the urinary bladder (PUB) is an exceptionally rare extra-adrenal location, accounting for only 0.05% of all bladder tumors and 1% of all pheochromocytomas, with only about 200 cases reported literature [3, 4]. Due to its rarity, the clinical presentation, diagnosis, and management of PUB remain challenging for clinicians.

The most common clinical manifestations of PUB include paroxysmal hypertension, headache, palpitations, and sweating, often precipitated by micturition or bladder manipulation. These symptoms are attributable to the intermittent release of catecholamines from the tumor. However, atypical presentations may occur, further complicating the diagnosis [2, 5].

Imaging studies, including computed tomography (CT) and magnetic resonance imaging (MRI), play a crucial role in the identification and localization of the tumor [3, 6]. Moreover, biochemical tests, such as plasma or urinary metanephrines and normetanephrines, are essential for confirming the diagnosis. In challenging cases, such as the one presented in this report, pathological examination is the only definitive way to confirm clinical suspicion of PUB [7].

Surgical resection remains the gold standard for the treatment of PUB, with transurethral resection or partial cystectomy being the preferred surgical approach, depending on tumor size and location. Preoperative management with alpha- and β-blockers is recommended to minimize the risk of intraoperative hypertensive crises and cardiovascular complications [8].

In this case report, we present a rare case of PUB in a 37-year-old patient, highlighting the diagnostic challenges, management strategies, and clinical outcomes. Our objective was to contribute to the existing literature on this rare entity.

Case Presentation

A 37-year-old man was referred to our center complaining of post-micturition palpitation, headache, and sweating. He had no history of hematuria or other irritative urinary symptoms. A detailed examination was performed to evaluate his symptoms.

An ultrasound of the urinary bladder revealed increased thickness of the right lateral wall. Further investigation with contrast-enhanced CT (CECT) demonstrated a suspicious mass in the right posterolateral wall of the urinary bladder (Fig. 1a, b). On admission, the patient's blood pressure was elevated at 175/70 mm Hg, and his pulse rate was 82 beats per minute and regular. No other remarkable findings were noted on physical examination.

Functional hormonal tests, including serum and 24-h urine catecholamine levels, were within normal limits. Other laboratory investigations, such as complete blood count, renal function tests, and liver function tests, were also unremarkable.

Cystoscopy revealed a small tumor, approximately 2 cm in size, located on the right posterolateral wall of the urinary bladder. The appearance of the tumor was distinct from that of typical urothelial tumors (Fig. 2a). Based on the clinical presentation and imaging findings, the patient was planned for open surgical enucleation of the suspected PUB.

The mass was successfully enucleated during the surgery, and the urinary bladder was sutured in two layers using absorbable sutures (Fig. 2b). The excised specimen was sent to the pathology department for histopathological examination. The pathologist confirmed the

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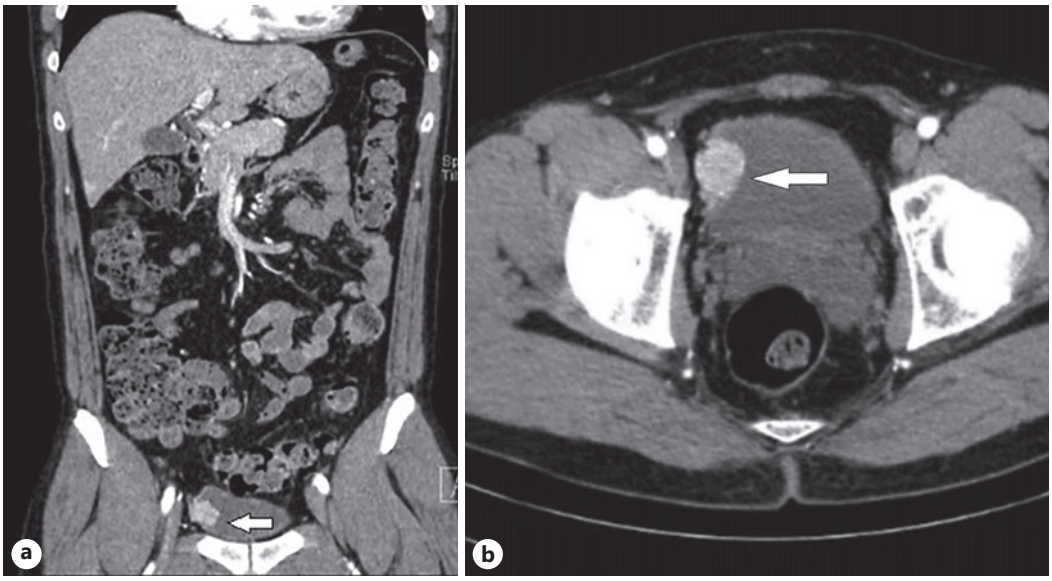

Fig. 1. a Coronal view of the abdomen and pelvic CECT shows urinary bladder tumor (white arrow). **b** Axial view of pelvic CECT shows urinary bladder tumor (white arrow).

diagnosis of pheochromocytoma of the urinary bladder (Fig. 3). The authors have completed the CARE Checklist for this case report which is attached as online supplementary material (for all online suppl. material, see https://doi.org/10.1159/000538073).

Discussion

PUB is an exceedingly rare entity, accounting for only 1% of all pheochromocytomas and 0.05% of all bladder tumors [1]. The clinical presentation of PUB can be diverse, and it may mimic other urinary conditions, making it challenging to diagnose. In our case, the patient presented with post-micturition palpitation, headache, and sweating, which are typical symptoms of catecholamine release.

It is noteworthy that the patient's functional hormonal tests, including serum and 24-h urine catecholamine levels, were within normal limits. This finding is unusual for pheochromocytomas, as elevated levels of metanephrines and normetanephrines are usually detected in these cases [2, 9]. The combination of the rarity of PUB and the lack of specific symptoms can render the diagnosis particularly challenging [10]. The normal hormonal test results in our patient emphasize the importance of maintaining a high index of suspicion and relying on a combination of clinical presentation, imaging, and cystoscopic findings for the diagnosis of PUB.

Imaging techniques, such as ultrasound and CECT, played a crucial role in identifying the suspicious mass in the right posterolateral wall of the urinary bladder. Although MRI can also be used for tumor localization, it was not performed in this case due to the clear findings on CECT. Cystoscopy provided further confirmation of the presence of the tumor, and its distinct appearance from typical urothelial tumors raised suspicion for PUB.

The surgical management of PUB presents unique challenges, primarily due to the potential for significant intraoperative catecholamine release [11]. Furthermore, the differential diagnosis of PUB can be particularly complex. The tumor might overlap with those of

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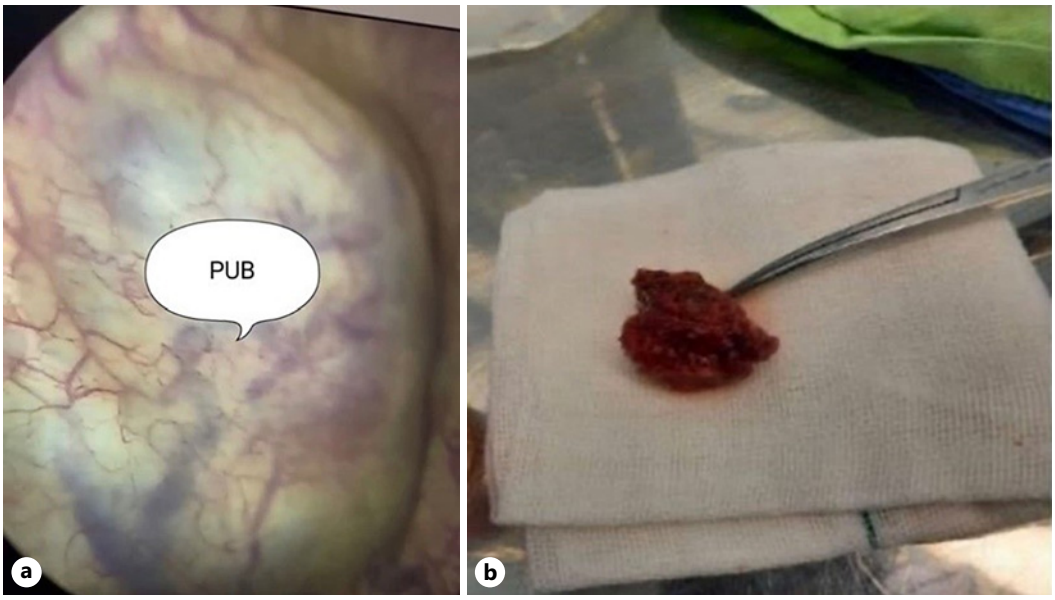

Fig. 2. a Cystoscopy view of the urinary bladder tumor. **b** Surgical resected specimen of the urinary bladder tumor.

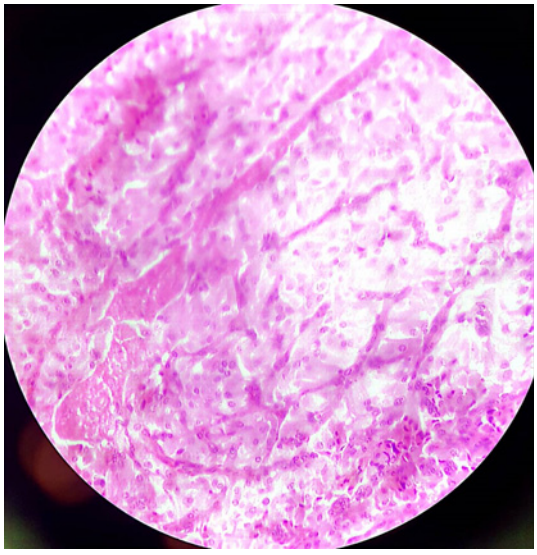

Fig. 3. Microscopic view of the urinary bladder tumor confirming paraganglioma of the bladder. Spindle or round tumor cells with basophilic, granular cytoplasm and round nucleus, vessels (hematoxylin and eosin staining).

more common urinary conditions such as urothelial carcinoma, requiring clinicians to consider a broad range of possibilities [12].

Surgical enucleation was successfully performed on our patient, and the pathologist confirmed the diagnosis of PUB. Surgical intervention is the mainstay of treatment for PUB, with transurethral resection or partial cystectomy being the preferred approach, depending on tumor size and location. In this case, the patient's blood pressure was well controlled preoperatively, and there were no reported intraoperative hypertensive crises or cardiovascular complications. These complications can be minimized by administering alpha- and β-blockers before surgery [8].

In conclusion, this case report highlights the importance of considering PUB as a differential diagnosis in patients presenting with post-micturition symptoms and paroxysmal hypertension. Early recognition and prompt surgical intervention are crucial for optimal management and favorable clinical outcomes. Furthermore, this case underscores the need for a multidisciplinary approach, including radiologists, urologists, and pathologists, in the evaluation and management of such rare and complex cases.

Statement of Ethics

Written informed consent was obtained from the patient for publication of the details of their medical case and any accompanying images and approval of the research protocol by an Institutional Reviewe Board. Ethical approval is not required for this study in accordance with local or national guidelines.

Conflict of Interest Statement

The authors declare no conflict of interest.

Funding Sources

This study was not supported by any sponsor or funder.

Author Contributions

A.Z. and M.B.: data curation; project administration; supervision; and writing – review and editing. F.M., B.N., and H.R.: data curation; investigation; writing – original draft; and writing – review and editing.

Data Availability Statement

The data that support the findings of this study are not publicly available due to privacy reasons but are available from the corresponding author upon reasonable request.

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