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Case Report

Management of bladder pheochromocytoma by transurethral resection

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Abstract Bladder pheochromocytoma is the most common extra-adrenal genitourinary tumor. Endoscopic management is feared due to the risk of intra-operative hypertensive crisis. We described a case of successful endoscopic management of a bladder pheochromocytoma and discussed its technical aspects.
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1. Introduction

Bladder pheochromocytomas are rare extra-adrenal tumors and are often recognized only at the time of transurethral resection (TUR) or as a histopathological surprise. If recognized preoperatively, these patients require a complete metabolic work-up, and a thorough search for any other metabolically active lesion in the body. The standard treatment for bladder pheochromocytoma is partial/radical cystectomy, since TUR is believed to induce intraoperative hypertensive crisis. Moreover, 10% of these lesions can be malignant and hence TUR alone may not suffice. Contrary to this notion there are reports of managing patients by TUR alone in select group of patients [1]. We reported a successful endoscopic management of a solitary bladder pheochromocytoma and highlighted a few technical tips to achieve complete endoscopic resection with minimal morbidity.

2. Case report

A 40-year-old male with bladder tumor was referred to us following an abandoned TUR due to intra-operative hypertensive crisis. On evaluation he gave a peculiar history of episodes of palpitation and dizziness at the time of

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micturition for 3 months. The patient also had one episode of hematuria, however there were no lower urinary tract symptoms. General physical examination and systemic examination were unremarkable. On further work-up urinary nor-metanephrine levels were raised (1255.2 μg/24 h; normal <659.5 μg/24 h). Computerized tomogram (CT) whole abdomen revealed a 3.8 cm × 4.3 cm polypoidal enhancing lobulated mass arising from the right postero-lateral wall of urinary bladder close to ureteric orifice (Fig. 1). A DOTANOC PET scan (GE healthcare, Chicago, United States of America [USA]) was done which corroborated the findings of contrast enhanced computerized tomogram (CECT) and did not reveal any other extravesical focus.

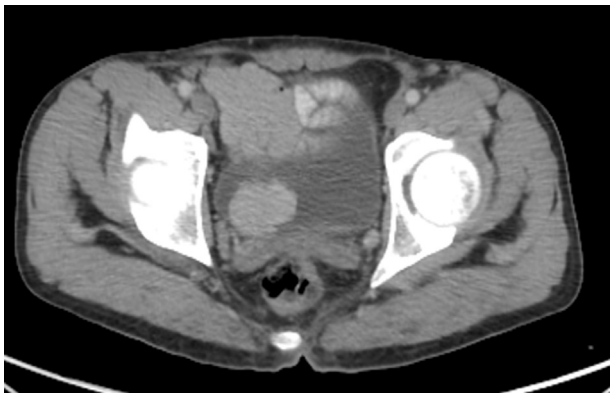

Figure 1 Computerized tomogram scan showing the right postero-lateral wall bladder mass.

The patient was optimized in consultation with endocrinologist on lines of extra-adrenal pheochromocytoma using sequential alpha-blockade followed by beta-blockade. In view of the lateral tumor location and proximity to ureteric orifice, the patient was planned for TUR under general anesthesia with invasive blood pressure monitoring, as it ensures better intraoperative hemodynamic management. Sodium nitroprusside and nitroglycerine infusion were prepared preoperatively for anticipated intraoperative hypertensive crisis. Cystoscopy revealed a sub-mucosally located tumor (Fig. 2A and B). Intraoperatively there was a rise in blood pressure on initial tumor manipulation which required use of parenteral antihypertensive medication only for a short duration. The bladder wall was circumferentially fulgurated at the base of tumor before starting the resection. Then the resection was done using short burst of current. To obtain a complete tumor clearance, the resection was done upto the perivesical fat (Fig. 2C and D). The entire procedure was completed uneventfully and patient's bladder was kept on irrigation for 1 day. Postoperatively his blood pressure remained normal without further use of alpha blockers. Check cystoscopy at 6 months follow-up showed no recurrence and a scar at the surgical site (Fig. 3). Repeat DOTANOC PET at 6 months follow-up showed no residual activity (Fig. 4A and B).

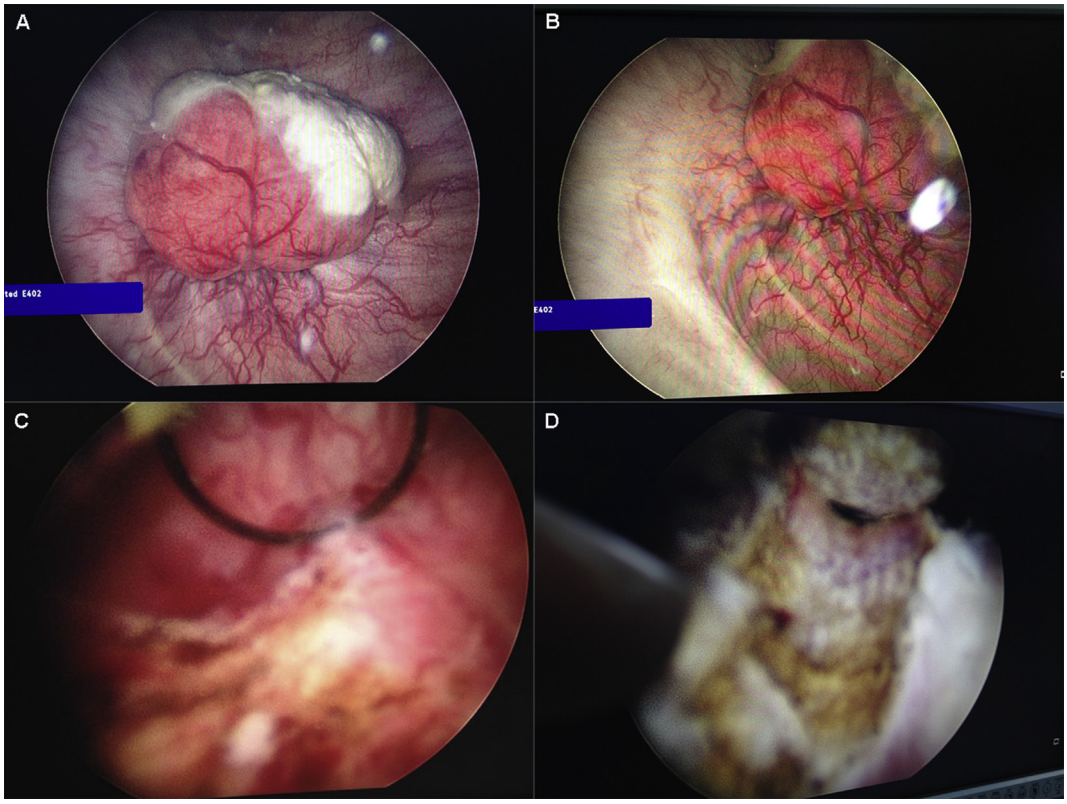

Figure 2 Cystoscopic view of bladder pheochomocytoma. (A) Intact mucosa with calcific plaque; (B) Lateral view showing the pedunculated tumor; (C) Transurethral resection using 26 Fr resectoscope and monopolar electrosurgical device; (D) Completed procedure showing dissection upto the perivesical fat.

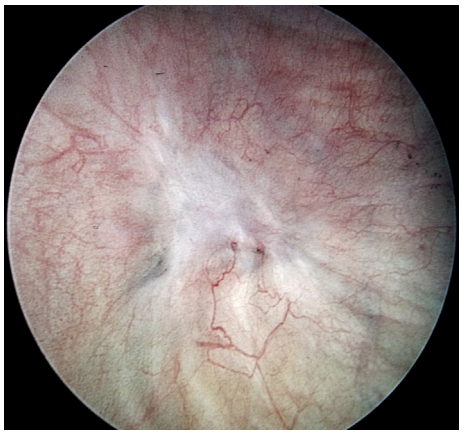

Figure 3 Cystoscopic view of resection site scar at 6 months.

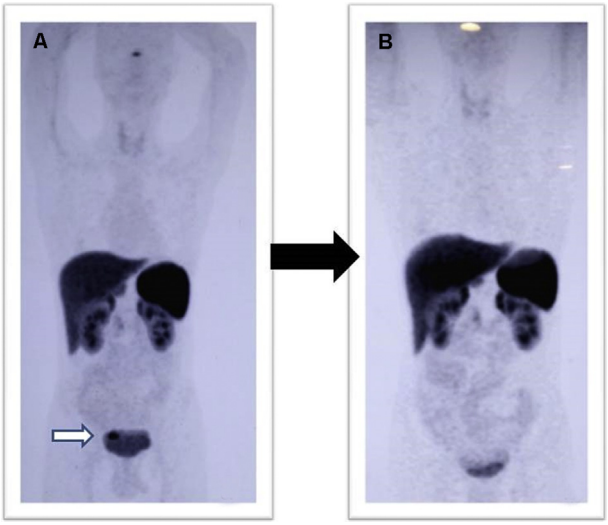

Figure 4 DOTANOC PET images (A) bladder pheochromocytoma (white arrow) and (B) showing complete resolution of tumor without any recurrence 3 months postprocedure.

3. Discussion

Bladder pheochromocytoma was first described by Zimmerman et al. in 1953 [2]. They are rare tumors which comprise less than 0.05% of all bladder tumors and less than 1.00% of all pheochromocytomas [3]. Among genitourinary tract, the urinary bladder is most commonly involved (79.2%) followed by urethra (12.7%), renal pelvis (4.9%) and ureter (3.2%) [3,4]. It involves patients in third to fifth decade and is usually submucosal or intramural with intact vesical epithelium. It commonly occurs in the trigonal region [5].

Ultrasonography is usually the screening modality which shows sharply circumscribed heterogeneously hypo-echoic lesions [6]. CT is widely available modality, providing high-resolution images in short time. Biphasic CT shows homogenous or heterogeneous hyper-enhancing soft tissue masses [5]. Calcification may be seen in about 10% of the cases as was seen in our case (Fig. 1A). Tumors are usually hypointense to iso-intense and show avid contrast enhancement [7].

Beilan et al. [7] have reviewed the literature and found the treatment modality of choice being partial cystectomy in almost 69% of cases. Most of the literature available is in form of case report and case series due to rarity of this entity. Similarly, in an earlier published case series from our centre the treatment of choice was advocated to be partial cystectomy citing reasons of hypertensive crisis intraoperatively [8].

Transurethral resection has been reported in handful of cases. However, in most patients the TURBT was done incidentally and the diagnosis was pathological and in hindsight [1,9]. In our case, diagnosis was evident preoperatively. The patient was optimized and electively prepared for surgery. Since it was a solitary primary lesion in the bladder and was close to the right ureteric orifice on the posterior bladder wall, decision was taken to manage the tumor endoscopically. Partial cystectomy was not feasible and would have added to the morbidity. With availability of better monitoring and effective perioperative anesthetic management, it is possible to manage the blood pressure fluctuations in these patients. Early coagulation of the feeding vessels to the tumor at its base and use of short burst of cutting limit the intraoperative blood pressure fluctuations. This may enable complete endoscopic resection of such lesions. The resection must be upto the perivesical fat to ensure completeness in order to decrease the risks of recurrence.

The role of functional imaging like MIBG (Siemens, Munich Germany) and DOTANOC PET is being defined in extra-adrenal pheochromocytomas. Level I evidence is being mounted in favor of DOTANOC PET over MIBG in terms of both sensitivity and specificity [10]. In our case the DOTANOC revealed the tumor clearly, ruled out other extra-adrenal lesions and also helped us in postoperative follow-up.

There is lack of high quality data and lack of organizational guidelines (*e.g.*, European Association of Urology, National Comprehensive Cancer Network and American Urology Association) thus a strict follow-up strategy is still to be defined [6]. A provisional follow-up strategy for functional tumors, regardless of stage has been formulated by Bielan et al. [7] which comprises monitoring of VMA, metanephrine and catecholamines levels 1 month post-surgery, then every 6 months for 2 years. In case of regional or metastatic disease axial imaging of the abdomen/pelvis should be performed every 3 month for 1 year, every 6 month for 1 year and annually thereafter for next 3 years [6]. Addition of functional imaging in form of DOTANOC PET as in our case can help in detecting the residual and recurrent disease at the earliest.

4. Conclusion

Endoscopic management of bladder pheochromocytoma is safe and feasible with proper preoperative optimization and use of specific endoscopic maneuvers in selected cases.

Author contributions

Study design: Aditya P Sharma, Ravimohan S Mavuduru, Shrawan K singh.

Data acquisition: Aditya P Sharma, Girdhar S bora, Bhagwant Mittal, Ravimoha S Mavuduru
Drafting of manuscript: Aditya P Sharma, Ravimohan S Mavuduru, Girdhar Bora, Shrawan K Singh.
Critical revision of the manuscript: Aditya P Sharma, Girdhar Bora, Ravimohan S Mavuduru, Vikas K. Panwar, Bhagwant Mittal, Shrawan K Singh.

Conflicts of interest

The authors declare no conflict of interest.

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