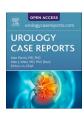
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Management of bladder tumors in pregnancy: A case of tumor prolapse and avulsion during labor

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ABSTRACT

Pregnancy presents unique obstacles to diagnosis and management of urologic disease. We present a case of a primigravid female with clot retention requiring evacuation in the operating room due to the avulsion of a bladder mass which prolapsed during labor. Tumor pathology demonstrated a low-grade spindle cell lesion positive for progesterone receptor (PR) and high mobility group A2 (HMGA2), suggestive of deep angiomyxoma versus a benign fibroepithelial polyp or inflammatory myofibroblastic tumor.

Introduction

Bladder tumors during pregnancy are uncommon, with few published cases. Diagnosis is difficult as the classic presentation of hematuria, irritative voiding symptoms, and outlet obstruction may be confounded by normal physiologic changes of pregnancy. Ultrasound, cystourethroscopy, and MRI are considered safe diagnostic modalities; however, any intervention requires careful consideration of pregnancy-related changes, such as bladder displacement and physiologic hydronephrosis. Due to the rarity of bladder tumors in this setting, specific management guidelines and indications for intervention have not been established.

Past case reports have highlighted successful tumor resection during pregnancy as well as concurrent partial cystectomy during delivery. We add to this body of literature, our presentation of a pregnant female with a small, asymptomatic bladder mass that had an unusual complication of avulsion and clot retention during labor and review the literature on considerations for intervention for bladder tumors during pregnancy.

Case presentation

A 34-year-old primigravid female was referred to urology after an incidental finding of a 3.34 cm bladder mass on 12-week prenatal ultrasound (Fig. 1). Flexible cystoscopy demonstrated a smooth, polypoid mass on a stalk, posterolateral to the right ureteral orifice, with

otherwise normal bladder mucosa and a normal urethra. Following discussion with the patient, the decision was made to postpone intervention until after delivery given the low likelihood of malignancy.³

The patient presented at term with vaginal bleeding and worsening hypertension and was admitted for labor. She stated she had been unable to void for several hours despite adequate fluid intake. Two hours following admission, she reported a tender, firm mass protruding from her urethra, and urology was consulted. Though the mass could not be manually reduced, a 14 Fr Foley catheter was placed, and the decision was made to reassess the mass after delivery.

During labor, the mass avulsed into the obstetrician's hand and was sent for pathological analysis. Immediately following delivery, the patient was noted to have gross hematuria, requiring the initiation of continuous bladder irrigation (CBI). Despite CBI, the patient was found to be in clot retention on post-partum day one, with symptomatic lightheadedness and a hemoglobin drop from 14.4 g/dL pre-partum to 7.9 g/dL. The patient was then brought to the operating room for cystoscopy with clot evacuation, which was notable for 200 mL of large, organized clot and bleeding arising from the apparent tumor bed. The base of the tumor was resected, and hemostasis established. After continued CBI overnight, the catheter was removed the following day. Postoperatively, her Hb stabilized after transfusion with 1 unit of packed red blood cells, and she was discharged on post-partum day 3.

The final pathology of both the avulsed tissue and resected specimen demonstrated mesenchymal tumor lined by benign urothelium (Fig. 2a).

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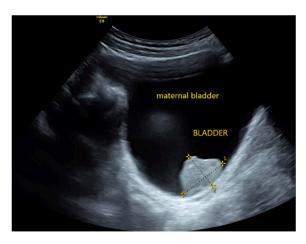


Fig. 1. Maternal bladder polyp on 12-week ultrasound (3.34 \times 2.46 cm).

The mass demonstrated moderate cellularity, prominent vasculature, and low grade spindled and stellate cell types in a fibromyxoid and hemorrhagic background (Fig. 2b). Immunohistochemical stains were positive for PR, factor 13a, and HMGA2 (Fig. 2c and d). While the mesenchymal origin was consistent with smooth appearance on initial cystoscopy as well as the demonstrated angiogenesis, a definitive diagnosis could not be made, with differential diagnosis including deep angiomyxoma, benign fibroepithelial polyp, and inflammatory myofibroblastic tumor.

Discussion

This case is interesting and important in several aspects. Bladder tumors during pregnancy are rare. In addition, the secondary presentation of urinary retention and ultimate tumor avulsion during labor have not been previously reported. This rare complication highlights a potential role for early surgical management. Lastly, the indeterminate pathology and hormone receptor positivity are noteworthy.

The rarity of bladder tumors during pregnancy has made it difficult to define an optimal treatment algorithm. If the tumor is felt to be benign, surveillance with regular prenatal ultrasounds may provide sufficient monitoring. However, this case demonstrated an

unanticipated consequence of the surveillance approach. We were unable to identify any previous publications of bladder tumor prolapse and avulsion during labor. Perhaps size of the lesion, its location within the bladder, and length of the stalk are all characteristics that contributed to this presentation.

In addition, the tumor's PR positivity on immunohistochemistry raises the possibility that the lesion was impacted by hormonal changes during pregnancy, making it a bigger risk for complications over time than it was at original presentation. The outcome therefore was unexpected and required urgent intervention.

In contrast to surveillance, recommendations exist to perform TURBT in pregnant women with noninvasive transitional cell carcinoma. In Spahn and colleagues' 27-case review of bladder tumors in pregnancy, including 4 low-risk bladder tumors managed with transurethral resection, no significant morbidities, fetal deaths, or tumor recurrences were noted. Additionally, timing of intervention remains an important factor. Even with considerations for fetal development and lung maturity, there may be opportunity throughout the second trimester as well as early in the third for elective resection with relatively little risk to the fetus. Nonetheless, intervention at any point during pregnancy includes the general risks of anesthesia, bleeding, infection, pre-term labor, fetal injury or loss.

The final pathology in this case was unusual as mesenchymal tumors represent only 5% of all bladder tumors. PR expression is feature of deep angiomyxomas, which are locally invasive and recur in half the cases regardless of size or margins. Although there are case reports describing recurrence of angiomyxomas in the setting of pregnancy, the role of pregnancy-related hormone fluctuations is not clear. In contrast, an inflammatory myofibroblastic tumor may be slow growing with little risk of metastasis, and a fibroepithelial polyp would be unlikely to recur. Due to the ambiguity, she will require close surveillance and possible repeat resection if recurrence is seen.

Conclusion

Optimal management of bladder tumors during pregnancy is not established given their rarity and limited reported experience in the literature. This case demonstrates the need for careful consideration of all treatment options during pregnancy, including early surgical intervention where appropriate. This may include patients with larger

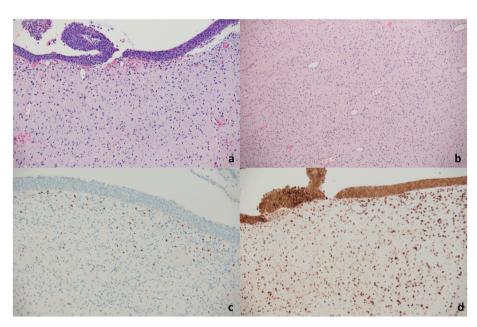


Fig. 2. Histopathology of bladder tumor. (a) Urothelial epithelium with lesion underlying. (b) Demonstration of hypercellularity and numerous blood vessels. (c) Focal PR positivity. (d) HMGA2 positivity.

tumors or those with tumors in locations that may make urinary retention, or possibly even the rare occurrence of prolapse or avulsion of the tumor, more likely.

Consent

Consent was obtained from the patient prior to procedure.

Declaration of competing interest

The authors have no conflicts of interest.

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