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Urethral Catheter-Related Bladder Wall Lesions Simulating Inflammatory Pseudotumor in a Neonate

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Manuscript Preparation E
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Conflict of interest: None declared

Patient: Male, Newborn
Final Diagnosis: Inflammatory pseudotumor of the bladder
Symptoms: Microscopic hematuria • oliguria • priapism
Medication: —
Clinical Procedure: Urinary catheter
Specialty: Urology

Objective: Unusual clinical course
Background: Inflammatory pseudotumors of the bladder are rare in newborns. Considering the potential for malignancy, invasive diagnostic assessment and therapeutic surgical excision have been described as the approach of choice.
Case Report: We present a case of urethral catheter-related injury causing multifocal lesions simulating inflammatory pseudotumors of the bladder, diagnosed in a newborn presenting with persistent priapism. This case is distinct by virtue of its very rapid spontaneous regression of the bladder wall lesions within 4 days.
Conclusions: Our neonatal case, along with previously described pediatric pseudotumor of the bladder, show the potential for spontaneous regression of these lesions and emphasizes the importance of balancing the potential risks and benefits of aggressive diagnostic or therapeutic interventions. Frequent diagnostic imaging and close follow-up should be considered as an alternative in young pediatric populations.

MeSH Keywords: Granuloma, Plasma Cell • Infant, Newborn • Urinary Bladder Neoplasms

Full-text PDF: <http://www.amjcaserep.com/abstract/index/idArt/891097>

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Background

Benign tumors of the bladder are uncommon in children. To date, there has been only 1 reported neonatal case [1]. This condition usually prompts invasive diagnostic and therapeutic approaches, including a biopsy, to ascertain its benign nature and exclude the potential for malignancy [2]. Here, we report a case of a neonatal mass-like lesion of the bladder possibly triggered by urinary catheter insertion, which is remarkable for its rapid regression during the 4 days after diagnosis.

Case Report

A male preterm neonate was born at 34⁺⁵ weeks of gestation with a birth weight of 2848 grams at a regional hospital, to a 33-year-old mother with a history of hypothyroidism, depression, and essential hypertension controlled by labetalol. She had an otherwise uneventful pregnancy, with 4 normal antenatal ultrasounds. The infant was born by uncomplicated spontaneous vaginal delivery, with Apgar scores of 9 and 9 at 1 and 5 minutes of life, respectively.

On first day of life, we noticed the neonate had persistent priapism, but the rest of the physical examination was otherwise unremarkable. The priapism was closely monitored and there was decreased urine output on the second day of life. A quick non-invasive bladder measurement revealed a residual volume of 70 cc. The instrument (BVI 3000 BladderScan[®]) does not provide an image sufficient to determine if a bladder mass is present. The medical team was not able to determine if there was an association between the priapism and the bladder retention. Multiple attempts to insert a 6 French urinary catheter were unsuccessful. Therefore, a 5 French nasogastric tube was inserted through the urethra and remained indwelling until the next day, when a voiding cystourethrogram (VCUG) was performed. The infant was immediately transferred to our tertiary-care Neonatal Intensive Care Unit (NICU) for further assessment and work-up.

On arrival at the NICU, the newborn had normal vital signs and physical examination but still had persistent priapism without macroscopic hematuria. His initial blood work showed blood urea nitrogen (BUN) of 2.0 mmol/L and creatinine of 73 μ mol/L, and his urine analysis revealed microscopic hematuria. The rest of his laboratory work-up was unremarkable, including a normal complete blood count. The lobulated posterior contour of the bladder on the VCUG called for an abdominal ultrasound, which revealed heterogeneous mass-like lesions, predominantly hyperechoic, measuring 2×1×3 cm, with lobulated well-defined margins, protruding from the floor and posterior wall of the bladder (Figure 1). There was also evidence of some vascular flow on the Doppler interrogation (Figure 2). Both kidneys

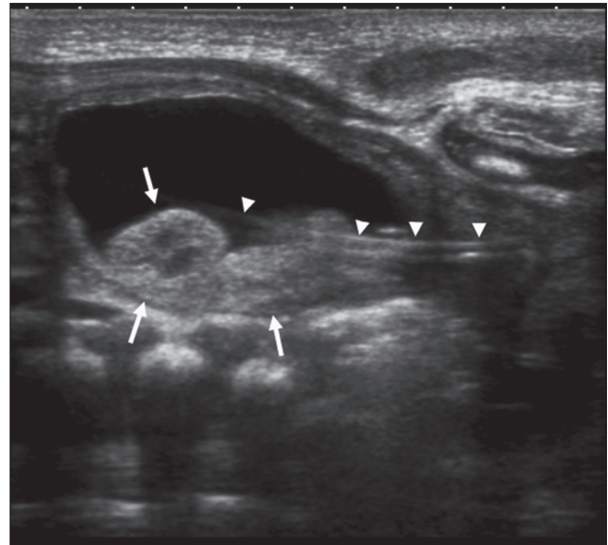


Figure 1. Ultrasound image in the mid sagittal plane shows the lobulated mass (arrows) and the catheter (arrowheads).

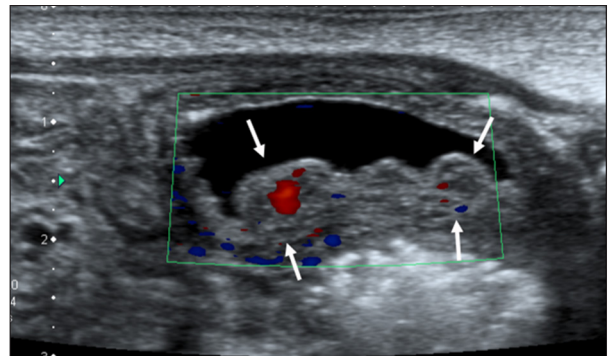


Figure 2. Color Doppler ultrasound shows the mass (arrows) with vascularity.

were normal in morphology and location. A pelvic MRI on the following day showed superficial lobulated submucosal lesions in the posterior wall and the floor of the bladder, with diffuse bladder wall thickening and irregularity. These lesions showed a homogeneous low/intermediate signal on T1 and high/intermediate signal on T2, without enhancement after the administration of gadolinium (Figures 3 and 4). The imaging features suggested an inflammatory pseudotumor of the bladder or a malignant tumor such as an embryonal rhabdomyosarcoma. Hematoma related to a blunt trauma induced by catheter urinary attempts was not considered because of the vascularity seen on ultrasound (Figure 2).

Following the initial investigations, and as per the current literature recommendation to investigate bladder wall mass, an invasive cystoscopy and biopsy was scheduled to establish the diagnosis. Considering the possibility of slow regression in inflammatory lesions of the bladder, a pelvic ultrasound

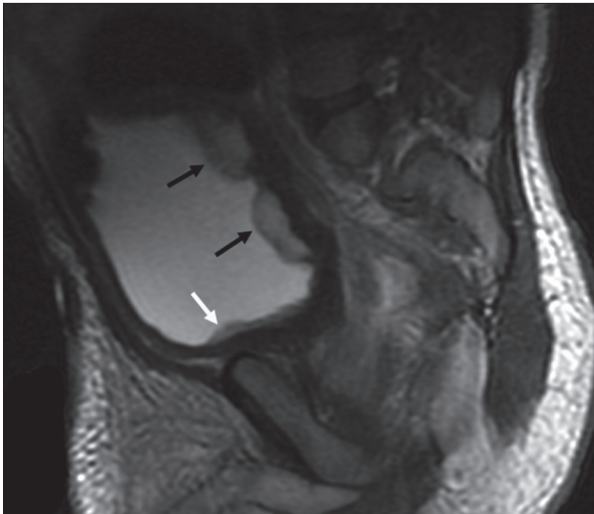


Figure 3. Sagittal T2 MRI (SE 2800/95) image of the bladder. Lobulated superficial hyperintense lesions extending from the posterior wall (black arrows) and the floor of the bladder (white arrow).

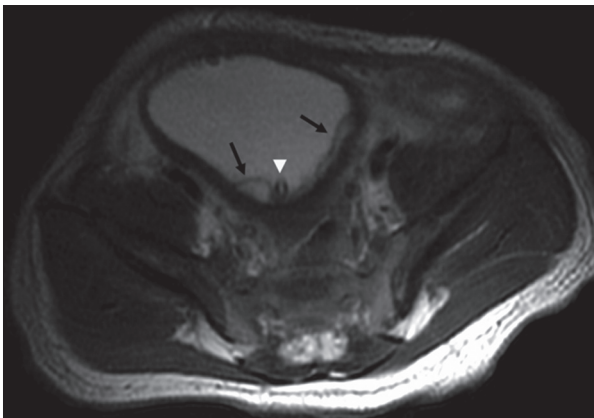


Figure 4. Transverse T2 MRI (SE 3416/85) image of the bladder. The hyperintense lesions (black arrows) adjacent to the catheter (arrowhead).

was performed before proceeding to invasive interventions. The repeat imaging, which was performed 4 days after the initial workup, showed complete resolution of the bladder mass with no evidence of bladder wall thickening or irregularity (Figure 5). The newborn genitourinary examination and voiding pattern at the time of the exam was also completely unremarkable. The cystoscopy was cancelled and a clinical diagnosis of inflammatory pseudotumor was considered based on the radiological findings and the complete regression of the masses. Of note, the Doppler ultrasound of the penis and the scrotum were both normal and the persistent priapism gradually resolved during the first 5 days of life. A follow-up bladder ultrasound examination at 1 year of age was completely unremarkable.

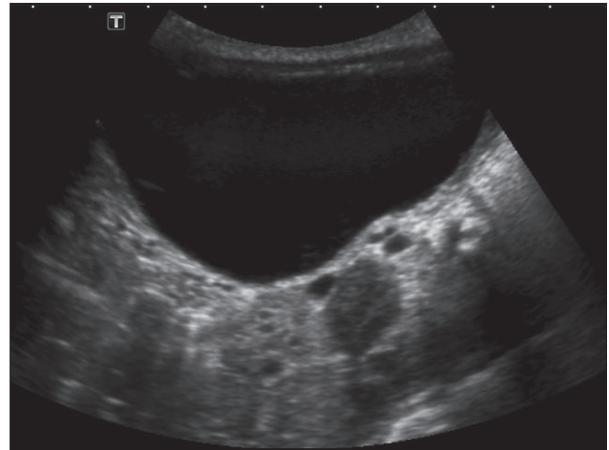


Figure 5. Ultrasound image in the mid sagittal plane on day 6 of life. No mass is visualized.

Discussion

Inflammatory pseudotumors of the bladder are rare proliferative lesions that are mainly described in middle-aged adults [3]. They are rare in the general population and extremely uncommon in infants. The etiology of bladder inflammatory pseudotumors remains unclear in the literature, although prior chronic inflammation, instrumentation, or surgery have been reported as potential predisposing factors in adults [4–6]. Although the bladder scan done prior of the urinary catheterizations did not provide detailed images of the bladder wall to confirm normal anatomy, an inflammatory reaction of the bladder wall caused by the multiple unsuccessful urinary catheterizations or the nasogastric tube insertion into the bladder was raised as a possibility in our case. To the best of our knowledge, a simple procedure such as urinary catheterization causing multifocal lesions mimicking pseudotumor of the bladder in neonates has never been described in the literature. Inflammatory reactions triggered by more invasive bladder instrumentation such as postoperative spindle cell nodule are distinct in their clinical characteristics and could not explain our case [3,7]. In our case, radiological characteristics and the very rapid regression of the lesions prompted us to suspect an inflammatory pseudotumor, possibly triggered by urinary catheter insertion, as the main clinical diagnosis. The presence of the priapism complicated our analysis. It is unlikely that the lesions as observed could have contributed of the persistence of the penile erection. Given that the priapism preceded the bladder catheter insertion, we assume that these 2 findings were distinct. Our investigation suggested an idiopathic etiology of the priapism, considering that secondary causes such as polycythemia, neurological conditions, and urinary obstruction secondary to posterior urethral valve were excluded [8].

The significance of inflammatory pseudotumors remains unknown in children and infants. Hematuria, dysuria, and frequency have been the most commonly reported presenting

symptoms [1,2,5,9–12]. The malignant potential of these tumors are yet to be determined, although so far there has been no report of metastasis of these lesions following surgical excision. The inflammatory pseudotumors and low-grade sarcomas of the bladder, although immuno-histologically distinct, overlap widely in clinical and radiological features [1–3,9,11–13]. Therefore, when iatrogenic causes cannot be excluded, invasive biopsy and surgical excision of the tumor has been the main diagnostic and therapeutic approaches of choice. The reports of inflammatory pseudotumors of the bladder in infants and children are limited to less than 50 cases in children and only 1 report in neonates [1]. The only report of conservative management of an inflammatory pseudotumor of the bladder was in a 6-year-old child who had 6 weeks of close observation followed by complete spontaneous regression [9]. Our case report, to the best of our knowledge, is the only report of bladder catheterization inducing a severe inflammatory reaction mimicking pseudotumor of bladder in a newborn. The potential for spontaneous regression of either mass-like injury to the bladder as described in our case, or inflammatory pseudotumor as described by Fletcher et al. [9] emphasizes the importance of balancing the need for aggressive intervention. The limitation of our case report is the inability to obtain a bladder biopsy that could have confirmed the histopathology, due to the quick, spontaneous, and complete regression of the bladder mass before the performance of the diagnostic cystoscopy.

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Conclusions

Considering the challenge clinicians face in differentiating inflammatory pseudotumors from the neoplasms of the urinary tract, and the burden of major surgeries in neonatal population, an initial conservative strategy with frequent imaging within a week after the initial diagnosis may be an appropriate alternative option for infants. Further steps may include confirmation of the diagnosis with cystoscopy and biopsy if there is progression or no signs of tumor regression. In case of improvement, weekly imaging follow-up of the patient should be considered so that full resolution of the tumor is ensured.

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Conflict of interest

Samira Samiee MD, Luis Guerra MD, Kaldhoun Koujok MD, Monica Rebollo-Polo, MD and Thierry Daboval, MD, MSc have no conflicts of interest to declare.