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Bladder mass in newborn: case report and review of literature

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ABSTRACT

Benign proliferative lesions of the bladder are exceptional in neonates. We describe a case of a 3 day-old neonate, presenting with bloody meconium and sonographic diagnosis of bladder mass. Cystoscopic biopsies were performed and a diagnosis of polypoid cystitis was made. The patient was treated conservatively and the lesion healed during follow-up. We review the literature of other cases of neonatal bladder masses.

INTRODUCTION

Benign proliferative lesions of the bladder are considered rare in children and exceptional in neonates. The presenting symptom of these lesions is sometimes haematuria but it may include urinary obstruction, inflammation or be completely asymptomatic. The diagnosis is not always straightforward, even by experienced pathologists (1), and in some cases the aetiology can remain unclear. We describe a 3 day-old female neonate with an polypoid cystitis of the bladder and review the literature.

CASE REPORT A full term 1 day-old female neonate was admitted at NICU of our Department presenting bloody meconium. Physical examination was unremarkable. Routine laboratory tests showed the following: RBC count 3.9×10^6 per mm^3 ; WBC count of 18,100 per mm^3 (N: 59%; L: 35%); Hb 14.5 g%; platelets 262,000 per mm^3 . Abdominal x-Ray was negative. At observation, the abdomen was flat and non-tender at palpation. A 6 Fr Foley catheter was put in for monitoring the diuresis. Samples of urine, blood and meconium were collected for culture and the patient was started on intravenous (iv) ampicillin and netilmicin. On day 3 of life, because of the persistence of rectal bleeding, an abdominal ultrasound was performed, which was negative except for a well vascularized pseudo-polypoid mass at the posterior wall of the urinary bladder measuring 7 mm. On day 4 of life, laboratory tests were repeated as follows: RBC 3.4×10^6 per mm^3 ; WBC count 43,300 per mm^3 (N: 59%; L: 35%); Hb 13.2g%; platelets 339,000 per mm^3 . Collection of three urine samples for cytology testing was started for three consecutive days. On day 7, urine and meconium culture were negative and urinary cytology displayed typical urothelial cells, several granulocytes and some red cells. A cystoscopy was performed, showing a rose, grape-like polypoid lesion between the posterior and superior wall of the urinary bladder (Fig. 2) and four endoscopic biopsies were performed. The urinary catheter was removed the day after the procedure. At histological examination, the samples had pseudopolypoid appearance. Oedema and acute inflammatory

infiltrate composed of neutrophilic granulocytes were evident in the lamina propria with exocytosis of the neutrophils from the venules (Fig. 3A and B). Reactive atypia of the overlying urothelium were also evidenced. The histological features were suggestive of polypoid cystitis. The blood culture documented a candida albicans infection and amphotericin B was started. After initiation of therapy for fungal infection, the patient's condition improved day by day and rectal bleeding disappeared. On day 14, the patient underwent a bladder ultrasonography, which showed that the mass had significantly reduced. On day 20, blood count was regular, ultrasound showed no sign of bladder lesion and the patient was discharged. Patient was followed-up at 1, 3 and 6 months with abdominal ultrasound, which were negative.

DISCUSSION

In the new-born period, bladder masses are extremely rare, including rhabdomyosarcomas (RMS) (2-4), inflammatory pseudotumor of the bladder (1), hemangioma (5), nonspecific/reactive lesion (6) and polypoid cystitis/inflammatory pseudopolyp (6). The neonatal presentation of RMS has an incidence of approximately 4 in 1,000,000 live births (7). The radiologic features of bladder RMS are not specific and benign mural masses of the bladder are difficult to differentiate from RMS (4). A precise diagnosis can demand additional clinical and pathologic examination. On the other hand, sometimes benign lesions of the bladder are often misdiagnosed as malignant lesion, even by experienced pathologists (1). In fact, inflammatory pseudotumor of the bladder, a benign reactive proliferative lesion of myofibroblasts (8), was misdiagnosed for sarcoma in four of 38 cases described in paediatric age that underwent total cystectomy or pelvic exenteration; two had chemotherapy and radiotherapy (1, 9). Just one case of inflammatory pseudotumor has been described in new-born. The etiology of this entity in the bladder remains unclear. Inflammatory pseudotumor has been associated with urinary tract infection, previous surgery or trauma but, in most patients no etiologic factor has been determined (1, 2, 9). Urinary bladder hemangioma should

be included in the differential diagnosis of intravesical polypoid mass on ultrasonography in children. Although hemangioma is a common benign tumor occurring in various parts of the body, it is a very rare primary tumor of the bladder. We could identify only a single reported case of bladder hemangioma in a new-born (5), which underwent cystoscopic biopsies. If a hemangioma is suspected, we propose avoiding endoscopic biopsy or resection because of the risk of bleeding, which can be very difficult to control through a small pediatric instrument. The new-born case reported here had a polypoid cystitis, only the second described case in English literature of in neonatal age (6). It was not sonographically or cystoscopically distinguishable with other localized lesions. All reported neonatal cases of bladder mass are summarized in Table 1. Differential diagnosis in the neonatal period with a reversible, reactive condition producing exophytic mucosal projection into the lumen of the bladder is impossible by imaging studies. In addition, blood and serologic tests are unremarkable for making a correct diagnosis. Pathological examination of a biopsy-specimen obtained endoscopically can allow a diagnosis even if cystoscopic biopsy in new-borns could be inconclusive because they are too superficial and thus cannot be considered a without-complication procedure. In cases of hemangioma, biopsy can result in a profuse, even fatal, bleeding (10, 11) or in a bladder perforation, as has been previously reported in a new-born with a RMS (3). Polypoid cystitis can be considered a pseudoneoplastic mimicker of a urinary bladder malignant lesion. This lesion, in older patients, is often described in association with indwelling catheter use and represents a non-specific mucosal reaction. The lesion grossly appears as friable, broad-based, often edematous polypoid or papillary projections, which can mimic a papillary neoplasm cystoscopically in adult (12). In our experience, such a lesion resembles a cluster of grapes, so that in a first instance, we considered a diagnosis of bladder rhabdomyosarcoma. Histologically, the classic examples show polypoid edematous and variably inflamed lamina propria, covered by reactive urothelium. The reactive and inflammatory nature of the process is considered key to distinction (12). Even if the histology was comfortable for a reactive lesion, the

specimens were not so deep to exclude an underlying rhabdomyosarcoma. For this reason, we performed a very short sonographic follow-up, considering an eventual subsequent open-biopsy. However, after 2 weeks the endoluminal lesion was significantly reduced and disappeared after 3 weeks. Sonographic behaviour was compatible with the pathological features so that the new-born was discharged, planning a short-medium term clinical and sonographic follow-up, that were uneventful. Unfortunately, we do not have any information about the other described case that has occurred in a new-born (6), so that we cannot know if a traumatic aetiology can also be supposed for that case. In conclusion, after a revision of the available literature, we can state that neonatal bladder masses are extremely rare, ranging from malignant tumor to absolutely benign lesion. In localized lesion, sonographic diagnosis is difficult and cystoscopic biopsy can be risky, especially if there is a suspicion of bladder hemangioma. Because some benign/reactive lesions can heal with a conservative management, open-biopsy or, worse, an aggressive surgery should not be considered as a first choice. If there was a vesical traumatic event, such as an indwelling catheter or urinary infection, a reactive lesion should be excluded as the number one cause of bladder formation. Moreover, even if we suggest a conservative or, at least, an endourological procedure, we believe that a short-medium term clinical and sonographic follow-up is wise.

REFERENCES

1. Asanuma H, Nakai H, Shishido S et al. Inflammatory pseudotumor of the bladder in neonates. *Int J Urol*. 2000;7:421-424.
2. Onal B, Ozdemir H, Arac M et al. Rhabdomyosarcoma of the prostate in a newborn: sonographic and CT findings. *Eur J Radiol*. 1995;21:106-108
3. Matsunaga GS, Shanberg AM, Rajpoot D. Prenatal ultrasonographic detection of bladder rhabdomyosarcoma. *J Urol*. 2003;169:1495-1496.
4. Esfahani SA, Montaser-Kouhsari L, Saeedi P et al. An antenatally diagnosed rhabdomyosarcoma of the bladder treated without extensive surgery. *Nat Rev Urol*. 2009;6:449-453.
5. Fernandes ET, Manivel JC, Reinberg Y. Hematuria in a newborn infant caused by bladder hemangioma. *Urology*. 1996;47:412-415.
6. Huppmann AR, Pawel BR. Polyps and masses of the pediatric urinary bladder: a 21-year pathology review. *Pediatr Dev Pathol*. 2011;14:438-444.
7. Lobe TE, Wiener ES, Hays DM et al. Neonatal rhabdomyosarcoma: the IRS experience. *J Pediatr Surg*. 1994;29:1167-1170
8. Freud E, Bilik R, Yaniv I et al. Inflammatory pseudotumor in childhood. A diagnostic and

therapeutic dilemma. Arch Surg. 1991;126:653-635

9. Fletcher SG, Galgano MT, Michalsky MP et al. Regression of inflammatory pseudotumor of the bladder in a child with medical management. Urology. 2007;69:e11-12.
10. Thomson HG, Burrows PE. Vascular malformation, in Cohen M and Goldwyn RM (eds): Mastery of Plastic and Reconstructive Surgery. Boston: Little, Brown, 1994, pp 352-373
11. Numanoğlu KV, Tatli D. A rare cause of hemorrhagic shock in children: bladder hemangioma. J Pediatr Surg. 2008;43:e1-3
12. Hameed O, Humphrey PA. Pseudoneoplastic mimics of prostate and bladder carcinomas. Arch Pathol Lab Med. 2010;134:427-443.

LEGEND

Video 1: Cystoscopy and endoscopic biopsy of the neonatal bladder mass.

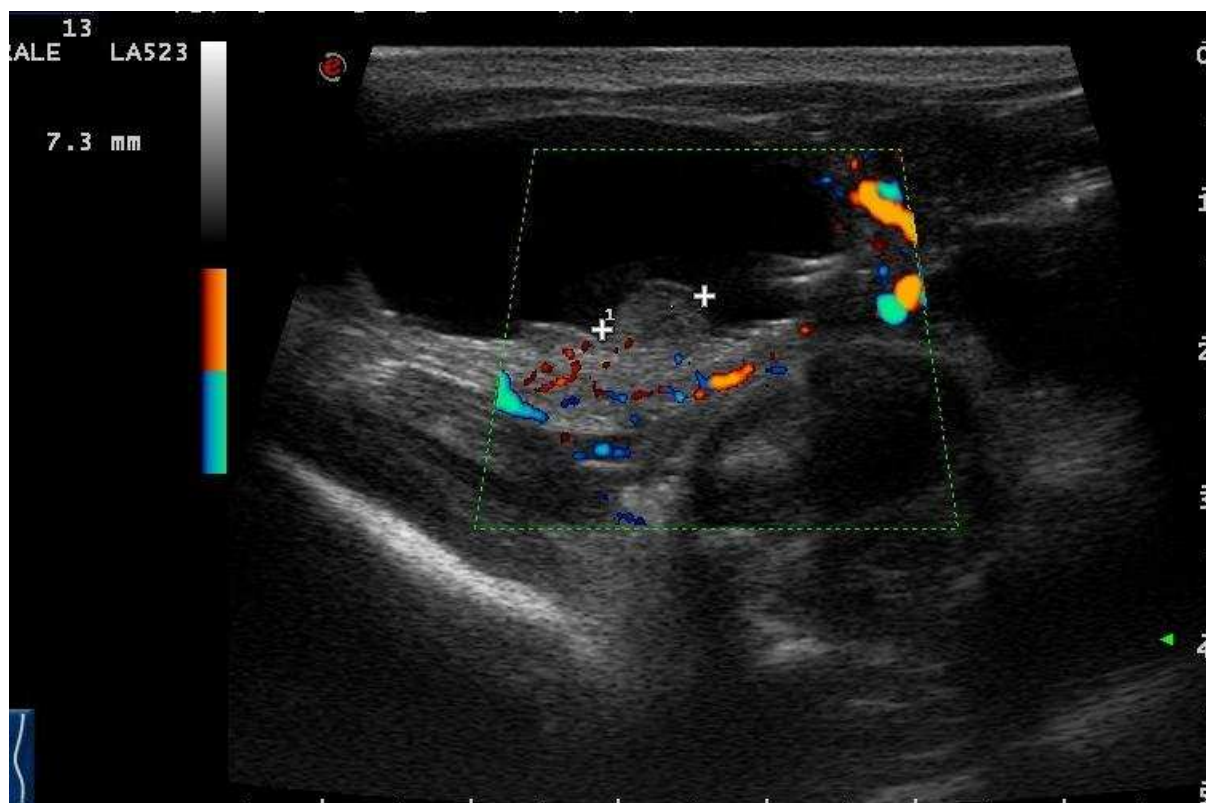
Figure 1: Bladder ultrasound shows a vascularized pseudo-polypoid mass at the posterior wall of urinary bladder measuring 7 mm (maximum diameter).

Figure 2: Cystoscopic view of neonatal bladder lesion.

Figure 3: Light microscopy. A) Exocytosis of Neutrophils in the lamina propria (Haematoxylin and eosin stain; original magnification, $\times 400$). B) Bladder mucosa showing oedema in the lamina propria and reactive changes in the urothelium (Haematoxylin and eosin stain; original magnification, $\times 200$).

Table 1: Reported cases of bladder mass in neonates are summarized.

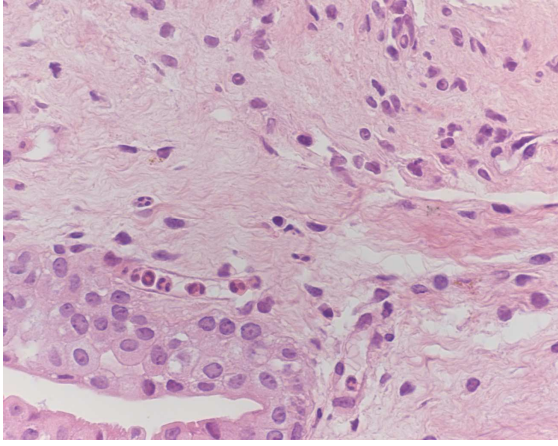
| Authors | Age of diagnosis | Diagnosis | Therapy |
|------------------------|-------------------------|--|----------------------|
| Huppmann AR, et al (6) | 11d | Polypoid cystitis / inflammatory pseudopolyp | Undefined |
| Huppmann AR, et al (6) | 0d | Nonspecific / reactive | Undefined |
| Fernandes ET et al (5) | 10d | Hemangioma | Cystoscopy / medical |
| Asanuma H et al (1) | 7d | Inflammatory pseudotumor | Open excision |
| Matsunaga et al (3) | Prenatally | Rhabdomyosarcoma | Partial cystectomy |
| Esfahani SA et al (4) | Prenatally | Rhabdomyosarcoma | Extensive surgery |
| Onal B et al (2) | 10d | Rhabdomyosarcoma | Open excision |



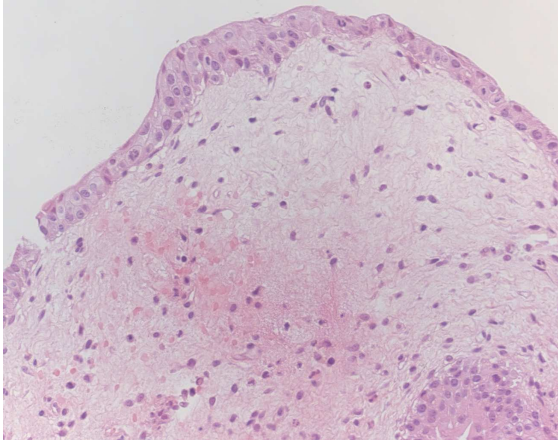
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