Case: Spontaneous bladder rupture presenting as sudden-onset abdominal pain in a child after many years in remission from bladder rhabdomyosarcoma

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Introduction

Bladder rupture in the absence of trauma (spontaneous bladder rupture) is a rare but life-threatening phenomenon, commonly associated with pelvic malignancy [1,2] and post-vaginal delivery[3]. In children, case reports of spontaneous bladder rupture are most often idiopathic or associated with congenital genitourinary malformations (e.g. posterior urethral valve), and bladder augmentation surgery[4]. Since bladder rupture often presents with acute abdominal pain, the diagnosis can be missed leading to delayed treatment and poor outcomes. Here, we describe the second case in the literature, to our knowledge, of spontaneous bladder rupture in a pediatric patient with prior bladder rhabdomyosarcoma.

Case report

An 11-year old male presented to a community emergency room with sudden onset abdominal pain that began after voiding. His past medical history is significant for bladder rhabdomyosarcoma diagnosed at age 4 and treated with partial cystectomy, chemotherapy (cyclophosphamide, actinomycin-D, and vincristine), and radiation as well as Grave's disease controlled by methimazole. His rhabdomyoscarcoma has been in remission for 6 years. Physical examination was significant for lower abdominal tenderness and diffuse peritonitis. Ultrasound and subsequent CT scan (Figure 1) revealed moderate ascites. Blood work and urinalysis were unremarkable except for serum creatinine and urea, which rose over from 68 to 204 μM and 5.4 to 8.9 μM, respectively, over 20 hours. A bladder rupture was suspected, and consistent with this, a cystogram (Figure 2) demonstrated extravasation of contrast into the pelvis. A foley catheter was inserted and the following morning, the patient was air-lifted to our institution.

On arrival, his vital signs were stable, but continued to display signs of peritonitis. He was taken to the operating room for laparotomy and bladder repair. Upon entering the peritoneum, approximately 400 mL of clear fluid was suctioned. A small

perforation was noted in the posterior aspect of the bladder dome around an area of heavy scarring that corresponded to the location of the previous rhadomyosarcoma. Leakage of instilled methylene blue confirmed the location of the tear.

The bladder was next opened along the midline and inspected for gross signs of tumour recurrence. No abnormalities were seen with the bladder lining, ureteric orifices, and bladder neck. The scar and bladder tissue surrounding the perforation were resected and sent for histology to confirm absence of malignant cells. Finally, the bladder and incision were closed in multiple layers

The patient was discharged on post-operative day 5 with an indwelling urethral catheter. He was seen in ambulatory clinic two days later for a follow-up cystogram (Figure 3), which demonstrated no bladder leakage. His catheter was removed. Pathology revealed a focus of chronic inflammation with infiltration of eosinophils**.** He was voiding well at 1 year follow-up with normal uroflow/PVR and no sonographic evidence of cancer recurrence. However, he complained of nocturia likely due to reduced bladder compliance from prior radiation.

Discussion

Spontaneous bladder rupture is a rare phenomenon, especially in the pediatric patient population. The majority of reported cases occurred in children with prior bladder outlet obstruction[5], bladder diverticulum[6], bladder augmentation surgery[4], and neurogenic bladder[7].

In this report, we described the case of an 11-year old boy with spontaneous bladder rupture 7 years after treatment for bladder rhabdomyosarcoma. To our knowledge, this is only the second reported case of bladder rupture in a pediatric patient following treatment for pelvic malignancy. The first being a case of a 12 year old girl also treated for bladder rhabdomyosarcoma at age 1 using the same regimen as our patient[8]. We suspected that the cause of rupture in our case could have either been cancer recurrence or long-term sequelae of the initial treatment. Spontaneous bladder rupture secondary to radiation therapy is better characterized in adults receiving treatment for cervical[1], prostate[2], and other pelvic cancers. Radiation cystitis is a known complication of pelvic radiation and can manifest many years after therapy as bladder fibrosis and atrophy[9]. These consequences likely weakened the bladder wall in conjunction with the effects of reduced bladder capacity and chemotherapy leading to rupture in our patient.

The pathology report in our case noted a focus of chronic inflammation with eosinophils, a finding not typically associated with radiation exposure. One case report has implicated eosinophilic cystitis in the spontaneous bladder rupture of a young child[10]. The absence of eosinophilia, hematuria, and voiding dysfunction make the clinical diagnosis of eosinophilic cystitis unlikely and the finding of chronic eosinophilic inflammation probably incidental in our patient[11].

The most challenging aspect of spontaneous bladder rupture is arguably making the initial diagnosis. Most cases of spontaneous bladder rupture present as acute abdominal pain, and can mimic other causes. Indeed, our patient was initially thought to have appendicitis and ascites was an incidental finding on ultrasound leading to the correct diagnosis. It is critical that bladder ruptures be managed quickly to prevent further complications like sepsis, fistula formation, and long-term bladder dysfunction. Given the number of case reports, we feel it is reasonable to perform CT scans in pediatric patients with significant prior urologic history presenting with acute abdominal pain when the history and physical exam do not point to a clear etiology. The diagnosis of bladder rupture is then confirmed by cystogram. Intraperitoneal ruptures are classically repaired by laparotomy, while uncomplicated extraperitoneal ruptures may resolve with insertion of a foley catheter[12].

The literature on bladder outcomes after remission in patients treated with partial cystectomy for rhabdomyosarcoma is not comprehensive. Yeung and colleagues[13] performed urodynamic assessment on 4 patients with abnormal voiding patterns after partial cystectomy for rhabdomyosarcoma and could only identify 1 patient with abnormal bladder compliance; however, the relevance of this finding to bladder rupture is unclear. We feel that with current evidence it is not possible to predict patients at risk of bladder rupture, but when patients with history of urologic malignancy and/or surgery present with acute abdominal pain, it is important to keep bladder rupture on the differential diagnosis.

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Fig. 1. Computed tomography scan demonstrating free intraperitoneal fluid. Clips from the partial cystectomy are visible in the coronal view.

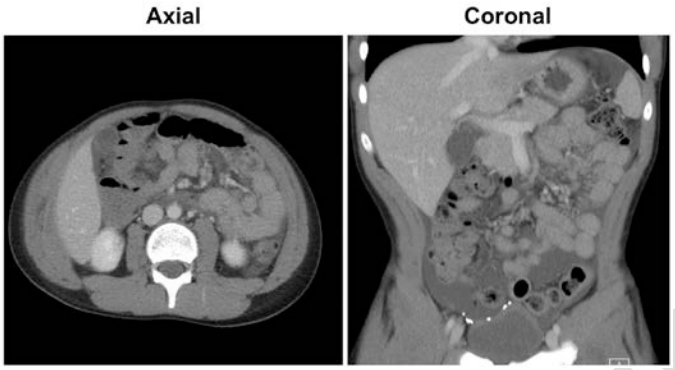

Fig. 2. Cystogram indicating marked pelvic extravasation of contrast as the bladder is drained.

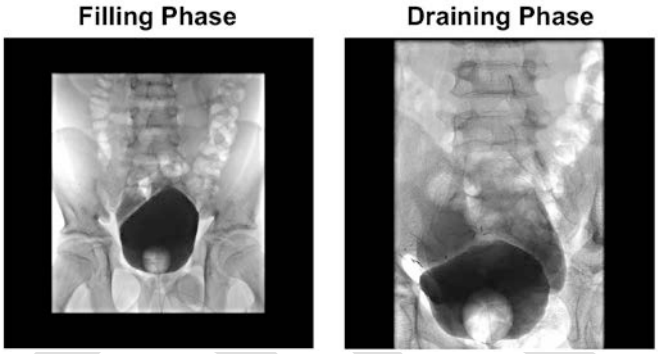

Fig. 3. Postoperative cystogram revealed no further leakage.