



Acquired posterior urethral diverticulum following surgery for anorectal malformations

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Abstract

Purpose: Despite significant advances in the surgical management of anorectal malformations (ARMs), many children still experience significant debilities from potentially avoidable complications. One complication, the posterior urethral diverticulum, may have untoward consequences if not recognized and treated.

Methods: A retrospective cohort review was undertaken of male patients who presented to us with persistent problems after being operated on elsewhere for ARM. Twenty-nine patients presented with a urethral diverticulum. Their charts were reviewed for the type of malformation, prior repair, presentation, treatment, and postoperative follow-up.

Results: Twenty-nine patients were identified that fit the criteria for this study. To date, 28 patients have been managed with reoperation. Urinary complaints were the most common presenting symptoms. All patients were repaired using a posterior sagittal approach. Pathology of the diverticulum in one patient revealed a well-differentiated mucinous adenocarcinoma.

Conclusion: The incidence of acquired posterior urethral diverticulum has decreased with the popularization of the posterior sagittal incision. There is a theoretical concern that the incidence may increase with the use of laparoscopy for the treatment of ARMs especially those where the fistula is below the peritoneal reflection. Once detected, the diverticulum should be excised.

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Despite significant advances in the surgical management of anorectal malformations (ARMs), many children still experience significant debilities from potentially avoidable complications with urologic sequelae [1,2]. The most common urologic complications occurring with the surgical management of ARMs are rectourinary fistulas (23.3%), bladder injuries (22.5%), urethral injuries (20.2%), posterior urethral diverticula (17.8%), injuries to the external genitalia

(14%), and ureteral injuries (0.8%) [1]. Urethral diverticulum in males without ARM may be congenital in origin [3–5] or may occur following trauma, urethral stricture, or prior urethroplasty [5,6].

Failure to mobilize the distal rectum adequately and creating a too distally located colostomy have been noted to contribute to the occurrence of urologic complications in ARMs [2]. In addition, a suboptimally done high-pressure distal colostogram may contribute to errors in diagnosis and nonrecognition of rectourethral fistulas [2]. Ligating the rectal stump at some distance from the urethra increases the

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Fig. 1 Voiding cystourethrogram showing large posterior urethral diverticula and clear communication to urethra.

risk of formation of a diverticulum [1,7]. A diverticulum develops from a retained part of the rectourethral fistula and balloons out as more urine is sequestered in the pouch-like structure (Fig. 1). This diverticulum that is colonic mucosa bathed by urine risks urinary dribbling, stone formation, and infection. Of concern is the potential for the development of cancer [8].

We sought to review the clinical manifestation, treatment offered, and outcome in patients who presented to our clinic with a posterior urethral diverticulum after surgery for ARMs.

1. Methods

A retrospective cohort review of 260 male patients who presented to our institution over a 22-year period (1982–2010) with persistent problems after being operated on for ARMs was performed. Thirty patients presented with symptoms suggestive of a urethral diverticulum or were found to have a posterior urethral diverticulum during evaluation for fecal incontinence. Patients with congenital urethral diverticulum were excluded from this study. The relevant information on the type of malformation, previous repair, clinical presentation, management, and postoperative follow-up was analyzed.

2. Results

Thirty patients had posterior urethral diverticulum during the study period. All were males who had been operated on previously for ARMs. Twenty-three patients (77%) had an abdominoperineal or sacroperineal pull-through as a form of definitive repair before presentation. The other patients had a posterior sagittal anorectoplasty (4) and laparoscopic-assisted pull-through (2). The nature of the surgery was not known in 1 patient. They presented 6 months to 24 years after the initial definitive surgery (median of 9 years).

The most common urinary symptom was daytime dribbling, and 13 patients (45%) presented with this symptom. Other urinary symptoms included recurrent urinary tract infections in 5 patients, passage of mucus through the urethra in 2 patients, pain with ejaculation in 2 patients, urinary stones in 1 patient, and 1 patient with adenocarcinoma of the diverticulum. Twenty-four patients (83%) were incontinent of feces at presentation. One patient who had a laparoscopy-assisted pull-through for a rectobulbar urethral fistula presented with a calcified pelvic mass (Fig. 2). The median time to presentation with symptoms was 3 years (range of 4 months to 6 years after the repair). In the asymptomatic group, in which the diverticulum was incidentally found, the median time to presentation was 10 years (range of 1–16 years).



Fig. 2 Magnetic resonance imaging of the pelvis showing calcified mass in the posterior urethral diverticulum in a patient after laparoscopy-assisted pull-through of a rectobulbar urethral fistula.

Twenty-nine patients have been managed by reoperation (1 patient was lost to follow-up after cystoscopy but before the diverticulum could be excised). Twenty-eight patients (96%) had the diverticulum excised via a posterior sagittal approach (Fig. 3); 1 patient had a posterior sagittal anorectoplasty without excision of the diverticulum. In 50% of cases, the diverticulum was larger than 5 cm; and it was giant in size (larger than the bladder) in 3 patients. The diverticulum, at surgery, was in the bulbar urethra in 8 patients (58%), prostatic-bulbar junction in 2 (17%), prostatic urethra in 2 (17%), and bladder neck in 1 (8%). The location of the diverticulum was not stated in the other patients. In 7 patients (23%), there was difficulty catheterizing the patient at surgery, as the catheter kept going into the diverticulum.

One patient who was reoperated on at the age of 31 years had a malignant transformation of the posterior urethral diverticulum. The histology of the resected specimen was reported as a well-differentiated mucinous adenocarcinoma.

The patients have been followed up after reoperation for a time ranging from 1 to 26 years (median of 13 years). Of the 13 patients who presented with daytime dribbling, 10 patients (77%) no longer dribble urine, one is dry with a continent diversion, one was lost to follow-up before surgery could be done, and one is incontinent of urine. The patient that is incontinent of urine has an absent sacrum and was last seen 17 years ago after a 4-year follow-up. Eleven patients are continent of feces, 11 are clean on a daily enema, and 1 patient (who also has Crohn disease) has a permanent ileostomy. Nine patients of 22, who have attained puberty, have information regarding sexual function; 8 (89%) have a normal ejaculation; and one has erections but no ejaculation. Painful ejaculations reported preoperatively have resolved in the 2 patients who had the symptom.

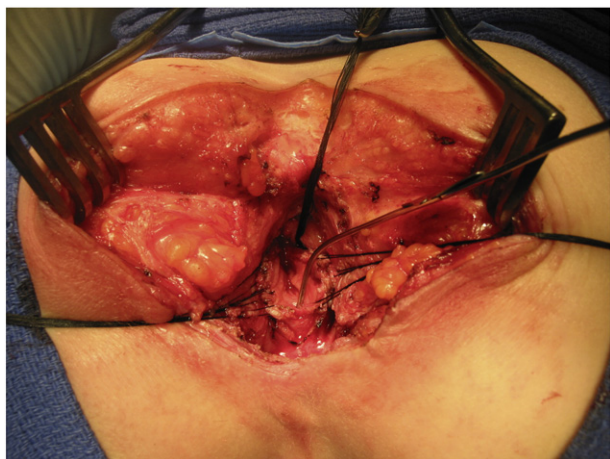


Fig. 3 An intraoperative photograph showing the posterior sagittal approach for management of the retained posterior urethral diverticulum. The anterior and posterior wall of the rectum is divided, and the diverticulum is below held open with stay sutures.

3. Discussion

Currarino [9] reported the first cases of acquired posterior urethral diverticula in a series of 3 children who had previously been operated on for ARM using abdominosacroperineal pull-throughs. Similarly, Vinnicombe et al [7] reported acquired posterior urethral diverticulum following abdominosacroperineal pull-through in 5 children with ARMs. Peña et al [10] had previously presented 20 cases of posterior urethral diverticula developing in children with rectourethral bulbar fistulas, all operated on transabdominally. In our study, 80% of the patients had an abdominoperineal or sacroperineal pull-through procedure. Suboptimal amputation of the rectum owing to inability to reach the fistula site is a major contributory factor in the development of the diverticulum.

The incidence of posterior urethral diverticulum appeared to have stagnated or declined in recent years as more surgeons became proficient in the posterior sagittal approach to repairing ARMs [1]. The advent of laparoscopy-assisted anorectal pull-through has heightened concerns about the development of posterior urethral diverticulum especially when the technique is used to treat cases of rectobulbar fistula [2]. The laparoscopic approach has been associated with posterior urethral diverticulum despite improved visualization from above and flush ligation of the fistula [11,12]. Koga et al [13] reported a case of posterior urethral diverticulum following laparoscopy-assisted pull-through in a boy who was treated for a rectourethral bulbar fistula. In a series of 9 children who had laparoscopy-assisted pull-through and were prospectively followed up until they were toilet trained, one developed a posterior urethral diverticulum [14]. Two patients in our study, with rectourethral bulbar fistula, developed posterior urethral diverticulum following laparoscopy-assisted pull-through of the rectum.

Methods used in the division of the rectourethral fistula during laparoscopy-assisted pull-through include the use of ties, sutures [15], harmonic scalpels (Ultracision, Ethicon), and clips [15]. Simple division of the fistula and placement of an indwelling urethral catheter, without using clips or sutures, have been proposed to lower the risk of injuries to the urethra [12]. However, one of 5 patients who had a simple division of the rectourethral fistula in that study developed a posterior urethral diverticulum at a follow-up of 10 to 19 months [12]. In addition, the presence of clips in a posterior urethral diverticulum resulted in calcification of a pelvic mass in one of our patients.

A technical modification to the fistula ligation during the laparoscopy-assisted approach for the treatment of “high anorectal malformations” has been suggested as a way of reducing the risk of leaving a segment of the fistula behind [11].

A child with posterior urethral diverticulum may be asymptomatic initially. An index of suspicion in a male child who had been operated on for ARM with a urethral fistula via an abdominal approach and now has urinary symptoms may

assist in the detection of patients that will benefit from further investigations. The major presenting symptoms in the present study were daytime dribbling of urine, recurrent urinary tract infections, urolithiasis, and passage of mucus through the urethra. This is similar to what had been reported by others [10,16]. The development of urinary tract infections and the calculi formation are because of stasis of urine in the diverticulum with secondary bacterial colonization [3]. Postmicturition dribbling occurs as the urine stored in the diverticulum drains out following completion of normal voiding. We also observed difficulty with catheterization in some patients (23%) owing to the presence of a false passage that is less angulated than the normal urethra [7]. A coude tip catheter was found useful to negotiate the urethra in such instances.

In such cases, imaging such as a pelvic magnetic resonance imaging with a Foley catheter in the rectum is an essential study (Fig. 1). Ultrasound may also be useful, as it will sometimes identify the diverticulum especially if filled by urine. A voiding cystourethrogram is a useful study to have because it will delineate the urethral anatomy; and a cystoscopy performed under anesthesia is also useful, although it could also be done in the setting of the definitive repair. The benefit of the cystoscopy for decision making regarding the excision of the diverticulum is apparent in the cases where the diverticulum attaches to the urethra with a narrow or stenotic neck. In these cases, the diverticulum could actually be much bigger than the imaging suggests.

The histology of resected posterior urethral diverticula typically shows colon-type mucosa [7,9]. The continuous exposure of this type of epithelium to the urinary stream has been suggested as a risk factor for malignant transformation [8]. The stump attached to the rectum in an untreated case of posterior urethral diverticulum has been associated with the development of adenocarcinoma [17]. One of the patients in our study had a malignant transformation of a long-standing diverticulum.

Although the treatment of acquired nonanorectal malformation related urethral diverticulum needs to be individualized and may be conservative [5], most authors advocate surgical excision for all cases of posterior urethral diverticulum [7,9,10]. The posterior sagittal approach was used to mobilize the rectum, resect the diverticula, and close the urethra in 20 patients with a satisfactory exposure [10]. Brain and Kiely [18] also used the posterior sagittal approach to resect a massive diverticulum in a child who had previously undergone an abdominoperineal pull-through for a "high anorectal malformation" and developed a posterior urethral diverticulum. Laparotomy has been used by some to achieve similar results [7,13]. Some authors, however, have suggested treatment of posterior urethral diverticulum only if associated with calculi or the diverticulum is massive [19,20]. All the patients in our study except one had the diverticulum excised via the posterior sagittal

route. The single patient who had his diverticulum left intact was managed very early in the series before the decision to excise all the diverticula was made.

The postoperation urinary and sexual functions in our patients, at a median follow-up of 13 years, have been encouraging. Daytime dribbling of urine was present in 13 patients preoperatively, but this had resolved in 10 patients (77%) postoperatively. We believe that the dribbling resolved after resection of the diverticulum and the loss of the "sequestration effect" of the diverticulum.

There is relative paucity of information on the long term follow-up of sexual function in patients who had been managed for posterior urethral diverticula. However, this study showed that 89% of the patients (8 of 9) have a normal ejaculation in the long term. This is predicated on not damaging the ejaculatory structures at both the first and subsequent surgeries. One must be careful with the reoperative dissection, as the seminal vesicles are closely attached to the portion of the urethra that involves the diverticulum.

In conclusion, posterior urethral diverticulum is a largely avoidable complication of surgery for ARMs. A meticulous preoperative preparation with an adequate distal colostogram and optimal ligation of the rectal stump will help in its prevention. The advent of laparoscopy-assisted repair of ARMs will lead to a resurgence of the problem if not judiciously used. The laparoscopic approach appears to be a risk factor for the development of a diverticulum when used for a rectobulbar urethral fistula or a no fistula defect which the rectum is at the bulbar level [21]. The PSARP approach is better able to delineate and dissect the distal rectum in such a case. We believe that laparoscopy should not be used for ARM with the rectum lower than the prostatic level. We recommend resection in all cases of posterior urethral diverticulum to prevent the possibility of malignant transformation and to avoid potential urinary symptoms such as daytime dribbling, infection, and stone formation.

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Discussion

Rebecca Meyers (moderator): Very nice presentation. I have a question for you. It seems you report a fairly high incidence of diverticuli. Has this incidence changed over time with the different types of operative repairs?

S. Alam: One of the reasons why we are seeing these numbers are [sic] that some patients were previously operated on in other countries and now are being adopted in the United States. Upon evaluation, we are finding an increased number of diverticuli. Again, the PSARP has really limited the number of patients who have this process.

Marc Levitt (Cincinnati, OH): If I could just add to that response, there is a large group of patients from the abdominoperineal era in whom this was basically an unknown complication for about a decade; and now we're seeing them in larger numbers again. I believe what is happening is more frequent use of a laparoscopic-assisted technique that uses a transabdominal approach to a rectum that lies below the peritoneal reflection, and that's the problem. If you approach the rectum with a high prostatic or bladder neck fistula laparoscopically, that is not a problem. However, in instances of low rectoprostatic or bulbar fistula below the peritoneal reflection, where the rectum and urinary structure s have almost a common wall, you're going to injure something or be more likely to leave a diverticulum because you're trying to get too close to the distal rectum. I think for low prostatic and bulbar fistulae, the PSARP is the better approach, and for a high prostatic and bladder neck fistula laparoscopy for that part of the dissection is the better approach. And that seems to be why we're seeing a spike in this complication.