URETHRAL DIVERTICULUM AFTER LAPAROSCOPICALLY-ASSISTED ANORECTAL PULL-THROUGH (LAARP) FOR ANORECTAL MALFORMATION: IS RESECTION OF THE DIVERTICULUM ALWAYS NECESSARY?

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Summary.- OBJECTIVES: With the increased use of minimally invasive surgery, the urethral diverticulum after anorectal surgery has become an issue. The few cases reported have been managed by surgical excision. We hereby report a case of urethral diverticulum after a laparoscopically-assisted anorectal pull-through (LAARP) procedure with a successful outcome after a period of active surveillance.

METHODS: A full-term boy who displayed a high anorectal malformation (ARM) and a recto-prostatic fistula underwent colostomy on the first day. He also showed associated malformations: bilateral low-grade reflux, horseshoe kidney and thoracic hemivertebrae; however, there were no signs of spinal cord tethering. Antimicrobial prophylaxis was started.

RESULTS: At the age of 3 months, he underwent a LAARP with a 3 abdominal-port approach. After complete dissection of the distal bowel, the recto-prostatic fistula was identified and tied with metallic clips. A 10 mm trocar was inserted through the centre of the sphincteric complex, which had been previously identified under laparoscopic view during perineal electrical stimulation. The anorectal pull-through was accomplished without tension. The bladder remained stented for 14 days. On the 18th postoperative day, a voiding cystourethrogram (VCUG) showed a 15 X 5 mm image of the diverticulum at the level of the membranous urethra. After 6 months, a new VCUG showed a normal urethra with neither signs of the diverticulum nor strictures; persistence of grade 2 reflux on the right side and resolution of the reflux on the left. When the boy was one year old his colostomy was closed uneventfully. Six months later he had not come into the emergency since the operation and voided with a normal flow.

CONCLUSION: This report suggests that LAARP is a feasible approach for ARM, although urethral diverticulum is a major concern. It may evolve without complications, and eventually resolve spontaneously. Active surveillance might be an option in selected asymptomatic patients; however a longer follow-up is advised to constitute better evidence supporting that policy.

INTRODUCTION

The repair of an anorectal malformation is a difficult surgical procedure and complications like urethral injuries may occur, especially with boys suffering from recto-urethral fistulae (1, 2). The risk of complications is significant due to the physical proximity of the urethra to the rectal pouch and also due to abnormal anatomy presented by the rectourethral fistulae (3). Awareness of the preoperative anatomy and surgical experience are fundamental to avoid unnecessary injuries (4).

After the report of Peña and Hong on patients with Anorectal Malformation (ARM) repaired thought posterior sagittal anorectoplasty (PSARP) who developed total continence in 25% of cases (5), Georgeson et al. invented the laparoscopic-assisted anorectal pull-through in order to avoid large incisions which might damage the sphincter muscle and nerves supply (6). Currently, laparoscopic-assisted anorectal pull-through (LAARP) is being proposed as a surgical option for male patients with a high-type imperforate anus and recto-urethral fistula (6, 7).

LAARP offers advantages over posterior sagittal anorectoplasty (PSARP), such as an excellent view of the rectal fistula, the creation of an accurate pull-through canal for the bowel, and minimally invasive abdominal and perineal wounds (6-9). With the laparoscopic stimulator the direct observation of the contraction of the puborectalis sling allows an evaluation of the functional contractility and an accurate colonic pull-through in the centre of the muscle complex (10).

There are few cases reported of urethral diverticulum after anorectal malformation repairs, and all of them have been managed by surgical excision (2, 4, and 11). We hereby report a case of urethral diverticulum after a LAARP with a successful outcome after a period of active surveillance.

METHODS

Description a case of a full-term boy who was born with a high anorectal malformation (ARM) underwent colostomy on the first day. The previous history was one of a controlled antenatal pregnancy without obstetric complications. The
study of associated malformations showed bilateral low-grade reflux, a horseshoe kidney and thoracic hemi-vertebrae. There were no signs of spinal cord tethering. Antimicrobial prophylaxis was started. At the age of two months a distal cologram diagnosed rectoprostatic fistula.

RESULTS

When the boy was three months old he underwent a LAARP with a 3 abdominal-port approach. Beforehand fistulae were identified under cystoscopy vision and a ureteric catheter was inserted through the fistulae to facilitate further management. After complete laparoscopic dissection of the distal bowel, the recto-prostatic fistula was identified, the ureteric catheter removed and it was tied with metallic clips.

The centre of the sphincteric complex was identified using perineal electrical stimulation, and under laparoscopic guidance a 10 mm trocar was inserted through it. The distal bowel was retrieved with a grasper and the anorectal pull-through was accomplished without tension.

Post-operative management was nil. Oral intake was started 48 hrs after surgery, and the bladder catheter with antibiotics remained for twelve days. He was discharged uneventfully seven days after surgery.

Two weeks later a dilatation program was started. On the 18th post-operative day, a voiding cystometrogram (VCG) showed a 15 X 5 mm diverticulum image at the level of the membranous urethra (Figures 1A – 1B), vesicoureteral reflux grade 2 on the right side and grade 1 on the left side. An ultrasonography showed non-dilatation. After 6 months there were no clinical problems and a new VCG showed a normal urethra with neither signs of the diverticulum nor strictures; persistence of grade 2 reflux on the right side and resolution of the reflux on the left (Figures 2A – 2B). When the boy was one year old the colostomy was closed.

FIGURE 1A y 1B. VCG on the 18th postoperative day showing a 15 X 5 mm diverticulum at the level of the membranous urethra.
uneventfully. After six months of follow-up after the last surgery, he has remained clinically well, has not come into the emergency department and voids with a normal flow. During this period he has not shown constipation or soling and he continues with urinary antibiotics prophylaxis until the next vesicoureteral reflux evaluation. A final cystoscopy was done at 18 months, which showed no signs of diverticulum.

**DISCUSSION**

Urethral diverticulum is a rare pathology, which may be acquired primarily or secondarily (12). Primary diverticula are a near 10% of all diverticula (12). For the other 90% the following may be recognized as acquired causes: urethral manipulation, surgical complications, instrumentations, artificial sphincter, isquemic events, and urethra compression in medular injury patients (12).

With children, urethral diverticulum can be seen after ARM repairs with a total incidence of 2% of cases after an abdominoperineal approach (2, 4). Few cases of urethral diverticulum have been found after LAARP (7). Children with this complication may present symptoms of micturition disturbance, urinary incontinence, urinary infections, obstructive uropathy, stone formation (13) and malignant transformation (2, 3, 14). That is why surgical excision of the diverticulum is always indicated.

The diverticulum may be the consequence of a long segment of rectum left attached to the urethra after the procedure (7) or of an inflammatory process produced by a reaction to the clips. The treatment advocated for urethral diverticulum is surgical excision through a posterior sagittal incision (4) or an abdominal approach (10).

Technical difficulties are more frequent with rectobulbar fistulae when the LAARP is used (7), because rectobulbar urethral fistulae are lower than rectoprostatic or rectobladder fistulae and the fistulae are more difficult to identify in all their extensions. This anatomic difference may leave a distal segment of fistulae if it is not completely dissected that will persist as a urethral diverticulum. Like Koga et al. proposed, we performed an intraoperative cystoscopy to identify the fistulae with a catheter which facilitated

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**FIGURE 2A y 2B.** New VCG 6 months later showing a normal urethra with neither signs of the diverticulum nor strictures; persistence of grade 2 reflux on the right-hand side and resolution of the reflux on the left.
the laparoscopic dissection (7). Other suggestions are to perform an intraoperative colonoscopy or use a 30 or 45-degree angle laparoscope. This technique could prevent injuries and complications to genitourinary structures.

This is the first case of urethral diverticulum after LAARP that has been managed conservatively. Previous reports of cases, almost all after PSARP, were treated with surgical excision, especially if they were symptomatic (11, 15). We opted for a conservative observational approach because the patient was asymptomatic, urethral dilatation didn’t show a rectal remaining attached to the urethra and no obstructive urinary signs were found.

Paediatric surgeons should suspect a posterior urethral diverticulum in any patient with urinary symptoms and a history of a repair via an abdomino-perineal approach (2). When urethral diverticulum evolves without complications, and no remainders of rectal pouches are found, it may eventually resolve spontaneously. Active surveillance might be an option in selected asymptomatic patients, however a longer follow-up is advised to constitute better evidence supporting that policy.

CONCLUSION

This report suggests that LAARP is a feasible approach for ARM, although urethral diverticulum is a major concern. It may evolve without complications, and eventually resolve spontaneously. Active surveillance might be an option in selected asymptomatic patients, however a longer follow-up is advised to constitute better evidence supporting that policy.

REFERENCES AND RECOMMENDED READINGS

(*of special interest, **of outstanding interest)