CASE REPORT

Two staged modified substitution urethroplasty using appendix-free flap

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SUMMARY

Reconstruction of long posterior urethra defect is technically challenging. Substitution urethroplasty is used in long, complex, recurrent posterior urethral strictures. This article presents a modified technique and the clinical outcome of two-stage substitution urethroplasty with appendix free flap and microvascular anastomosis. A three-year-old boy with a 5 cm iatrogenic posterior urethral defect was managed by urethral substitution using the appendix. An appendix-free flap was used according to anatomic limitations, employing the transposed inferior epigastric artery and saphenous vein to maintain conduit blood supply. The conduit was buried in the scrotum for 3 weeks and its viability monitored until the final reconstructive stage. Two-year follow-up with ultrasound and cystoscopy revealed satisfactory results. A well-vascularised bed and flap are the mainstays of substitution urethroplasty, so we suggest inferior epigastric artery perineal transposition and staged reconstruction as alternatives that may improve the blood supply of the neourethra.

BACKGROUND

Pelvic fracture is the most common cause of posterior urethral stricture in all ages, but it may occur due to congenital causes, infection or iatrogenic trauma in the paediatric group.¹ There is still a technical challenge in posterior urethra reconstruction, which mostly depends on the gap length, but an open surgical approach is considered the best method for management in these cases.²

While perineal anastomosis urethroplasty may be able to bridge a gap of up to 4 cm, urethral substitution or transpubic urethroplasty may be needed in larger gaps.³ Substitution urethroplasty is considered in treatment of long, complex, recurrent posterior urethral strictures, especially after several failed explorations and repairs.³

Unfortunately, the results of urethral substitution are not encouraging and recurrent stricture, anastomotic stenosis, diverticula and stone formation have been reported.² Well vascularised conduit and tension-free anastomosis are the main factors that account for functional results, while the bed of the conduit is often fibrotic and avascular in recurrent complicated strictures.⁴

The colon, jejunum and appendix have been used as pedicled flaps in reconstruction of the posterior urethra.⁵ There are several reports of acceptable results in substitution of the posterior urethra with vermiform appendix on its vascular pedicle.^{3 6} This article presents a modified technique and clinical outcome of two-stage substitution urethroplasty

with appendix-free flap and microvascular anastomosis. This modified technique may be a helpful alternative in those cases where the pedicled appendix does not reach the perineum, without applying tension on its vascular pedicle.

CASE PRESENTATION

A 3-year-old boy was referred after iatrogenic long posterior urethral stricture, multiple perineal explorations and failed reconstructive attempts. Posterior urethral trauma had occurred during a perineal surgery 18 months before referral. A cystostomy had been performed for urinary drainage and the boy's parents were frustrated as the boy was also physically and psychologically impaired.

INVESTIGATIONS

Radiological assessments (retrograde urethrography with simultaneous cystography via cystostomy) revealed a 5 cm gap in the posterior urethra from the penoscrotal junction up to the bladder neck (figure 1).

TREATMENT

Considering the previous failed attempts of transpubic urethroplasty, penile fasciocutaneous flap urethroplasty and also buccal mucosal graft urethroplasty and severe perineal fibrosis, we decided to perform a urethral substitution using the appendix as the conduit; however, while exploring the abdomen, the appendix pedicled graft did not easily reach the perineum, despite complete releasing of the meso-appendix and cutting of the ileocolic artery. The vascular pedicle was almost 5 cm in length and 3 mm in diameter, and severe tension on fine appendiceal vessels made it ischaemic while transposing it to the perineum. We converted the technique to a free appendix graft with microvascular anastomosis using the inferior epigastric artery and proximal saphenous vein to provide blood supply to the appendix free graft (figure 2).

Appendix exploration was performed via a transverse lower abdominal incision and the inferior epigastric artery was released in the rectus abdominis through the same approach. The inferior epigastric artery was cut and transposed to the groin through the inguinal canal. The medial accessory saphenous vein was also prepared for venous supply (figure 2). The vessels were washed out with heparinised saline after cutting off of native blood supply to the appendix, and end to end microvascular anastomosis was performed on the prepared vessels using 7-0 Prolene (video 1).

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Figure 1 (Left) Perineal scar due to multiple failed urethral reconstructions. (Right) Retrograde urethrography with simultaneous cystography shows a 5 cm posterior urethral gap.

We believe that a well-vascularised graft has a major role in the outcome of urethral reconstruction, so we decided to retain the appendix in the scrotum without any tension on the new vascular pedicle and allow it to become stable, and then reassess the viability before any further re-exploration of the perineum for final urethral reconstruction. The proximal end of the appendix was anastomosed to the scrotal skin as a stoma, to drain the appendiceal mucosal secretions and also for monitoring of conduit viability (figure 2).

Low dose heparin was also started during the operation and continued for 5 days in the postoperative hospital stay. Urethral reconstruction was planned 3 weeks later, through a perineal dissection, and after full excision of the stenotic fibrotic segment. The proximal urethra was defined by cystoscopic stenting via the cystostomy and a guide wire was passed through the cystoscope into the proximal urethra and the conduit, and through the distal urethra. End-to-end anastomosis was performed to bridge the urethral gap, using the well-vascularised appendix pedicled flap. An 8F silicon catheter passed through the neourethra on a guide wire, which had been placed retrogradely via the cystostomy before urethral reconstruction (figure 2).

Outcome and follow-up

The catheter and cystostomy drain were kept in place for 3 weeks before being removed.

Postoperative voiding cystourethrogram was performed a month later and revealed mild distal anastomotic stricture, which was managed by urethrosopic urethrotomy. The patient was able to void normally during our 2 years of follow-up. Urinary tract ultrasound and cystoscopic evaluation were performed intermittently during follow-up visits every 3 months. Mild proximal dilation was observed during follow-up, although the voiding calibre and patterns were acceptable. We performed cystoscopic urethrotomy at the anastomosis rings during the first three cystoscopic evaluations. The appendix mucosa became almost atrophic in the final stages of cystoscopic assessments.

During the last visit, the patient and his family were happy, and the boy was able to urinate voluntarily with good calibre and without hesitancy or dribbling (video 2). Final ultrasound evaluation was also normal, without bladder wall thickness or hydronephrosis, and final cystoscopy did not reveal significant stricture or bladder trabeculation. The patient is still in

Figure 2 (A) Schematic view of vascular reconstruction for free-appendix flap. (B) Microvascular anastomosis for urethral conduit. (C) Buried appendiceal conduit in the scrotum. (D) Well-vascularised appendiceal conduit in place. (E) Urethral reconstruction.

Video 1 Pulsation of appendicular vessels after microvascular anastomosis.

our follow up programme while both the patient and his family are happy, having resumed a normal life.

Discussion

Posterior urethral loss in children may occur due to trauma, severe inflammations or congenital reasons.⁷

Reconstruction of an extensive posterior urethral gap is always challenging, but the procedure has continued to evolve in the past few decades. The most challenging patients are those with a very long defect that does not permit primary anastomosis, and especially in those recurrent cases with multiple previous failed attempts and severe perineal fibrosis. The fibrotic bed does not provide a good scaffold for skin or mucosal grafts, and common techniques such as tabularised full thickness skin grafts are disappointing in these patients.³ On the other hand, parents and the patient are always frustrated and hopeless due to unsuccessful previous experiences, and seek a definite therapeutic intervention.

Substitution urethroplasty is advised for long gap recurrent posterior urethral loss, though the results are not encouraging, so consultation with the parents would be problematic.²

Provision of a well-vascularised conduit for urethral substitution may be performed using the colon, jejunum, ileum or appendix on the mesenteric pedicle.^{3 6 8}

Small or large bowel conduits need reductive tailoring, and complications such as diverticula formation and urethral stone or stricture are common, while the appendix seems a more compatible conduit for the urethra.³

Sometimes, providing a well-vascularised pedicled appendix flap may not be demanding because of anatomic variations or limitations. Free intestinal or appendix flap with micro vascular

anastomosis to the femoral artery and vein have been reported previously as an alternative in complex posterior urethral substitution.^{7 9}

Reviewing the previous experiences revealed a high rate of stricture after urethral substitution that necessitate frequent long-term probing or cystoscopic urethrotomies. It appears that poor blood supply is the main problem with neourethra, which may cause long-term complications such as recurrent stricture. We believe that radical excision of perineal fibrotic scar tissue is a critical first step in successful reconstruction. The second most important factor is blood supply, which may be compromised when there is a short vascular pedicle and tension on the flap pedicle. Even severe local oedema after extensive perineal dissection or vascular exploration in free flaps may compromise the conduit's blood supply.

Considering the fine appendicular vascular pedicle in our patient, we released the inferior epigastric artery and transposed it to the perineum to protect the appendix arterial pedicle from tension or compression at the time of anastomosis, instead of using the conventional method and anastomosis deep in the femoral artery. We decided to perform two-stage reconstructive surgery to allow the neourethra to become stable and well vascularised, so we buried the appendix free flap in a subcutaneous scrotal pouch without any tension placed on its new vascular pedicle and continued to monitor its viability by observing its mucosa at the stoma site, and finally performed the urethral substitution with minimal dissection. The result was encouraging during the long-term follow-up.

Learning points

- Urethral substitution may be the only choice in reconstruction of long gap complex and recurrent posterior urethral loss or stricture.
- The appendix appears to be a promising conduit to replace the missed urethra, and may be used either pedicled or free flap with microvascular anastomosis.
- We believe that a well-vascularised bed and flap, and a stable, well nourished viable conduit have a major role in long-term results of urethral substitution.
- We suggest inferior epigastric artery perineal transposition and staged reconstruction may achieve a promising blood supply for neourethras.

Contributors MH is the chief surgeon and performed the surgery, RS is corresponding author, followed the case, gathered data and prepared the manuscript. PSS was the consultant surgeon and also followed the case for a long period of time.

Competing interests None declared.

Patient consent Obtained.

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Video 2 Patient voiding in the follow-up.

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