CASE REPORT

Repair of Perineal Urethrostomy Stenosis Using Buccal Mucosal Graft in a Patient Diagnosed with Body Dysmorphia and Who Previously Underwent Total Penectomy, Bilateral Orchiectomy, and Scrotectomy: A Case Report

Patricia Anne Tagle, MD; Raul Carlo C. Andutan, MD, FPUA and
Mark Joseph J. Abalajon, MD, FPUA, FPhilGURS

Department of Urology, East Avenue Medical Center

Body dysmorphia is a debilitating disorder that centralizes on a preoccupation with one's physical appearance. Often, these individuals seek surgical correction in an effort to subdue this preoccupation. A majority of complications from feminizing gender reassignment surgery, consists of urethral stricture or stenosis, leading to voiding dysfunction. The patient is 39-year old male who underwent bilateral nipple removal, bilateral orchiectomy, scrotectomy and total penectomy with perineal urethrostomy, one year prior to consult. The patient eventually presented with acute urinary retention secondary to perineal urethrostomy stenosis. Urethroplasty with revision of perineal urethrostomy site using a buccal graft was done; and on follow up, he was noted to have good urine flow on uroflowmetry with mild lower urinary tract symptoms. Complex urethral strictures may be noted in patients with prior reconstructive history and lengthy areas of fibrosis. Although perineal urethrostomy is a valid surgical course of treatment for patients with complex strictures, improper technique, suboptimal patient factors, and, poor healing may lead to stenosis. The study aims to describe the use of a buccal graft as a viable alternative and easily reproducible technique to augment a revision perineal urethrostomy and lessen the recurrence of stenosis.

Key words: Buccal mucosal graft, reconstructive Urology, body dysmorphia, gender reassignment surgery

Introduction

The Diagnostic and Statistic Manual of Mental Disorders characterizes body dysmorphic disorder (BDD) as an impairing preoccupation with a perceived defect or flaw in the personal appearance. Patients often perceive this to look unattractive or deformed with increasing severity leading to poorer functioning and quality of life. These serve the foundation of its four criteria: A) Preoccupation with a perceived defect or flam, B) Repetitive behaviors or mental acts in response to the appearance concerns, C) Significant distress or impairment from this preoccupation, and D) Preoccupation not explained by body fat or weight.[1] In the United States, point prevalence was noted to be greater (2.4%) than other countries (1.7 - 1.8%).[1] Furthermore, studies done in Europe and North America have reported equivocal findings regarding gender differences.[2] A majority of patients have been noted to undergo surgery (dermatological, cosmetic surgery, and/or maxillofacial surgery) to remedy their preoccupation.[1-3] Prior to surgery, psychiatric evaluation and clearance is secured from two different psychiatrists explicitly defining this as the treatment of choice. Unfortunately,

surgical treatment was rarely found to improve overall symptoms with a majority expressing dissatisfaction on follow up.[2,3,4] As such, suicidal ideation and attempts are markedly elevated in this population.[1,2]

Feminizing gender reassignment surgery (GRS) as a whole includes psychotherapy, hormonal therapy, and a series of genital and non-genital surgical procedures more commonly indicated for patients affected by gender dysphoria[5] as opposed to body dysmorphia. The patient is also started on at least 12 months of hormonal therapy with estrogens and anti-androgens and required to undergo one year of social integration as the desired gender.[6] Although previous studies have noted urethral stricture and wound healing disorders to be the most frequent findings on follow-up[6], more recent studies have noted a dramatic decrease in these complications attributable to sufficient spatulation of the remaining bulbar and penile urethra.[5-7] A recent meta-analysis of complications for feminizing GRS noted a prevalence of 32.5% for overall surgical complications with urethral stenosis or stricture serving as the majority of these cases (14%). Despite this common complication, there was a noted disparity in its prevalence and correlation with surgical management. This may be attributable to a majority of reviewed cases attended to by non-urologists and thus lacking appropriate assessment and experience in managing lower urinary tract dysfunctions.[8]

Complex urethral strictures, as seen in failed reconstructions, often present with a challenging repair. In comparison to simple strictures, they often require extensive pelvic surgery due to: 1) a long area of fibrosis, 2) strictures associated with diverticulas, false passages and fistulas, or 3) extensive sphincter damage.[9] Apart from a history of surgery, cases often also include multiple urethral instrumentation which further complicates selection of the appropriate intervention.[10] Common interventions include skin flaps, buccal mucosal graft (BMG) and perineal urethrostomy.[11] Of the three, perineal urethrostomy creation is preferred for long strictures located in the proximal urethra and as an alternative to complex staged repairs.[12] Although the incidence of perineal urethrostomy stenosis post reconstruction has been equivocal, most studies concur that this outcome is often noted in patients with a history of multiple surgical procedures (i.e prior urethroplasty)[11-14] possibly attributable to the tissue's compromised blood supply.[14] We are presenting a case of a complex stricture with a history of surgical reconstruction and multiple urethral instrumentation and who underwent perineal urethrostomy repair with a buccal mucosal graft.

The Case

The patient in this case is a 39 year old male who sought consult due to acute urinary retention. One year prior, the patient underwent bilateral nipple removal, bilateral orchiectomy, scrotectomy and total penectomy with perineal urethrostomy. He had been diagnosed with body dysmorphia and maintained that he identifies as male with no desire to feminize his physical appearance or change this gender identity. The patient expressed feeling severely anxious and often perceived his genitals as tumors that did not belong in his anatomy. He then proceeded to undergo the aforementioned surgery with a non-urologist. No hormonal therapy was noted before or after this procedure. Additionally, although the patient's consent was taken, there were no noted clearances or evaluation from psychiatric services.

Post-operatively, the patient developed progressively worsening lower urinary tract symptoms. This prompted intermittent consult with multiple urologists for further management. He underwent at least two sessions of direct vision internal urethrotomy (DVIU) with minimal improvement. Due to the persistence of symptoms, he was then advised to start clean, intermittent, self-catheterization. Patient was initially compliant with this form of treatment; however, his symptoms exacerbated and ultimately led to acute urinary retention. On further urological assessment, the perineal urethrostomy site was noted to be completely stenotic (Figure 1). Prior to definitive surgery, emergency suprapubic cystostomy (STC) was performed to relieve the urinary retention.

Surgical Technique

The patient was placed in a lithotomy position under general anesthesia. The surgical site was

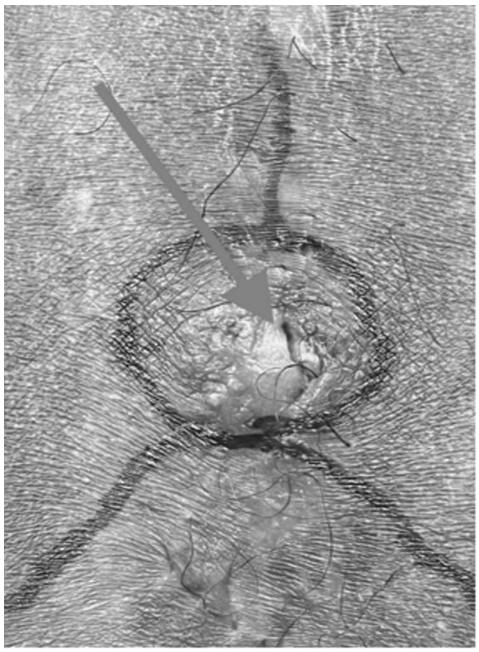

Figure 1. Stenosis of perineal urethrostomy

marked accordingly prior to being prepped with a betadinized solution (Figure 2a). A circumferential incision was done around the perineal urethrostomy site before being extended to an "inverted U" incision slightly inferior to the urethrostomy (Figure 2b). Upon further dissection, there were noted fibrotic portions of the bulbar urethra about 1.5cm from the urethrostomy site. These were carefully excised (Figure 3a) before the remaining bulbar urethra was mobilized proximally (Figure 3b) and pulled to the level of the skin incision.

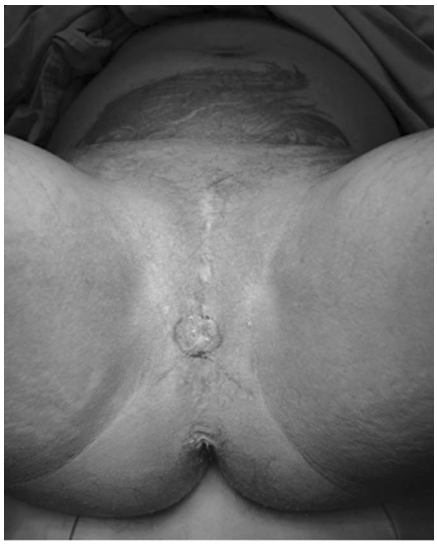

Figure 2a. Preoperative markings

Figure 2b. Skin incision

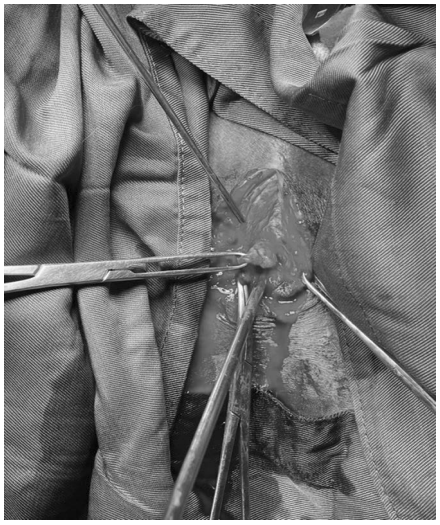

Figure 3a. Excision of fibrotic urethral segments

Figure 3b. Urethral mobilization

Intraoperative flexible cystoscopy (Figure 4) was done for complete assessment of the urethra. This revealed 95% obliteration of the urethrostomy lumen with a severely fibrotic bulbar urethra extending from the urethrostomy site to the membranous urethra approximately 1.0 cm from the sphincter. The distal urethra was then spatulated dorsally (Figure 5) while its ventral portion was carefully mobilized. The oral cavity was prepped and subsequently harvested for a buccal mucosal graft. The triangular shaped graft was approximately 1.5cm in width and 2.5cm in length and was quilted dorsally to the corpora cavernosa with vicryl 5-0 sutures (Figure 6). The apex of the perineal skin and "inverted U" incision were meticulously anastomosed to the ventral urethra (Figure 7) prior to closure. Finally, a siliconized Fr14 catheter was inserted with bolster dressing placed on the reconstructed area.

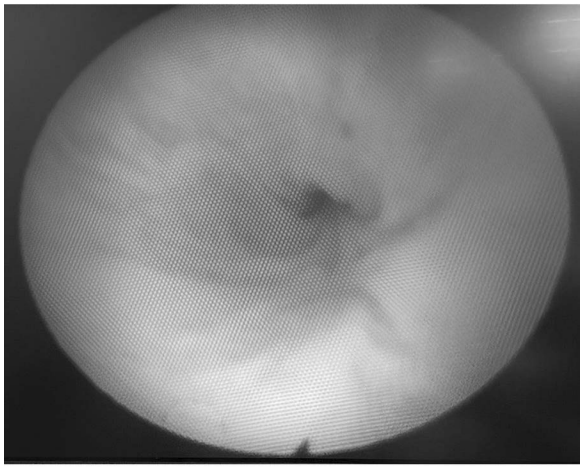

Figure 4. Intraoperative flexible cystoscopy of nembranous urethra

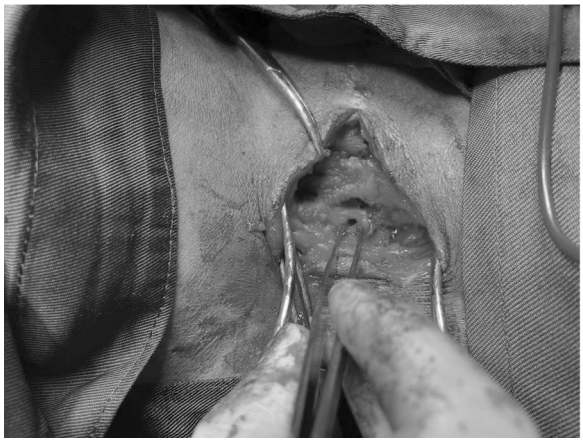

Figure 5. Supple urethra identified and spatulated dorsally

Figure 6. Buccal graft laid dorsally and quilted to the corpora cavernosa

Figure 7. Apex of perineal skin and inverted U incision anastomosed to ventral urethra

Results

Patient had an unremarkable postoperative course. Five days postoperatively (Figure 8), there was noted good wound healing at the reconstructed urethrostomy site and adequate output thru the indwelling catheter. After removing the catheter, patient was able to void freely with no straining or difficulty. Good graft take was also noted on his follow up 1 month post-surgery (Figure 9) with tolerable, mild lower urinary tract symptoms, based on IPSS, on follow-up one and three months post-surgery. Patient additionally underwent uroflometry, six months post-operatively with a Qmax of 18ml/sec.

Figure 8. Five days post revision

Figure 9. One month post revision

Discussion

BDD falls under the spectrum of obsessive-compulsive disorders and may manifest as repetitive behavior or mental acts in relation to the concerned physical appearance that ultimately causes clinically significant distress or impairment. Although studies have been mostly equivocal on gender distribution, larger reports done have found either an equal distribution between genders or a slight female predilection. BDD reported in men tend to have a preponderance for genital or muscular preoccupation.[1,3] An important distinction made in the most recent Diagnostic and Statistical Manual of Mental Disorders (2013) was its difference from gender dysphoria; unlike this, there is no conceived incongruence between the individual's expressed gender and their anatomical gender.[1] Patients often have a substantial impairment with daily functioning (social, occupation, etc.) and overall markedly low quality of life.[1,2] Thus, a majority of patients seek and receive surgical treatment for their apparent "flaws".[2] In a study done by Lai and colleagues (2010) examining the hospital records of cosmetic surgery patients over a three year period, 85.7% of patients with BDD were diagnosed during the preoperative workup for their planned cosmetic operations.[15] As in the present case, the patient has distressing preoccupation and severe aversion towards his nipples and genitalia; ultimately opting to undergo surgical management for specific removal of these parts only. The procedure done is essentially the penectomy component of feminizing GRS and as such, shares a similar complication profile due to two main issues: urethral angulation and stenosis secondary to devascularization.[16] This may manifest in deflected or slow voiding patterns, incomplete emptying, frequency, dysuria and spraying, which may contribute to the progression of dysphoria and be non-affirming.[16]

During this first surgical stage of GRS, the bulbar urethra is exposed and retracted prior to dissection of the vaginal space.[6] The urethra is then shortened by dividing the urethra in the proximal bulb and suturing the urothelium directly to the skin.[5] When the urethral meatus is positioned too distally or performed without spatulation, the urethra becomes angulated upwards towards the clitoris.[16] Although this method has a low rate of bleeding from the anastomosis, it may lead to antegrade micturition due to the inability to direct the urinary stream downwards.[5] Another common outcome is the high incidence of urethral meatal stenosis that is often due to the lack of spatulation,

poor mucosa to skin opposition or tissue ischemia from extensive mobilization.[8,16]

During the patient's first surgery performed by a non-urologist, the bulbar urethra was exposed and shortened, but was not spatulated, before being sutured directly to the skin during the creation of a perineal urethrostomy. Performing sufficient spatulation of the remaining bulbar and penile urethra to avoid stenosis has been considered the cause for the dramatic decrease in complications. From an incidence rate of up to 40% in previous literature, new series have reported a 2.9% incidence with only one person requiring revision.[5-7]

A number of techniques are available for the repair of perineal urethrostomy stenosis. Similar to other complex strictures, skin flaps or buccal mucosal grafts may additionally be utilized to increase the success of repair. An important key point to factor in repair for this case is the patient's history of surgery. Most use a scrotal skin flap due to its proximity to the involved area and because it may be used as either a vascularized flap or as a free skin graft.[5] However, the patient presented with no scrotum or excess skin available for an advancement flap. Furthermore, his history of previous repair may have caused a disruption in the blood supply and subsequently induced scarring, ultimately reducing the quality of the tissue involved.[11,12] Supplementation and/or repair with a flap alone may not be enough to ensure successful perineal urethrostomy because of its unreliable blood supply.[12] Sasam and Abalajon[14] described the use of two BMG's as dorsal and ventral inlays to augment the urethra in a female patient. Knowing the applicability of BMG in repairs of very proximal strictures, the decision was made to use a buccal graft to augment the revision of the perineal urethrostomy. The graft acts as a healthy substitution and upon onlay, provides a wide diameter neomeatus.[11] This allows an increased success when used in conjunction with perineal urethrostomy[11-13] with or without the additional use of skin flaps.[11,12]

Conclusion

Complex strictures are challenging because of their compromised blood supply, attributable to the history of surgical reconstruction. In the present case, the use of a buccal graft as well as sufficient spatulation of the remaining urethra allowed better post-operative outcomes. Thus in patients who have previously undergone a perineal urethrostomy, the use of a buccal graft to augment a revision perineal urethrostomy, is an easily reproducible technique, that can lessen the recurrence of stenosis. In the future, the authors recommend a larger case series with a longer follow-up period to fully evaluate outcomes for this particular technique.

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