

Recurrent Female Urethral Stricture Managed by Double Buccal Mucosa Graft Urethroplasty: Case Report

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Received July 01st, 2020; Revised July 12th, 2020; Accepted July 14th, 2020

ABSTRACT

Female urethral strictures (FUS) are relatively rare in urological practice but on occurrence, cause profound morbidity. No proven guidelines for diagnosis and management exist to direct clinical practice. Diagnosis is on the basis of clinical symptoms, a high index of suspicion, imaging, urethral calibration and cystourethroscopy. Urethral dilation and urethroplasty are the mainstay modalities of surgical management. Dilation often coupled with clean intermittent catheterization (CIC), though a first-line approach, lacks long-term success compared to urethroplasty. Novel techniques high lighting various reconstructive techniques estimate success rates; variably defined, as ranging from 57.1% to 100%. Given the lack of robust evidence for superiority of one technique over another, surgeon's experience is a key factor in procedure choice. We present M.W.G, a 58-year-old female who presented with obstructive urinary symptoms and frequent urinary tract infections (UTI) due to a recalcitrant FUS for nearly 2 years. She was diagnosed with Human Immuno deficiency viral (HIV) disease in 2015 and commenced on anti-retroviral medication (Dolutegravir/Lamivudine/Tenofovir). Voiding cystourethrogram (VCUG), urethral calibration, and cystourethroscopy confirmed the diagnosis. Double Buccal Mucosa Graft (BMG) urethroplasty was performed. Ultimately; she enjoyed normal unobstructed voiding during the entire 17 months of follow-up. BMG is a cost-effective option for FUS.

Keywords: Female urethral strictures, Urethral dilation, Vaginal flaps/grafts, Double buccal mucosa graft Urethroplasty

Abbreviations: FUS: Female Urethra Strictures; UTI: Urinary Tract Infection; PVR: Post-Void Residual; VCUG: Voiding Cystourethrogram; BMG: Buccal Mucosa Graft; CIC: Clean Intermittent Catheterization; AUASI: American Urological Association Symptom Index; AIDS: Acquired Immunodeficiency Syndrome; HIV: Human Immunodeficiency virus

INTRODUCTION

Although no universal definition of FUS exists, Osman et al defined FUS as 'A symptomatic, anatomical narrowing of the urethra based on failure of catheterization, urethral calibration, visual inspection, endoscopy or radiography' [1]. Smith et al. [2] suggest less than 14 F urethral caliber as the anatomical cut-off for consequential urethral narrowing. FUS unlike those in males, are relatively uncommon in urological practice with incidence largely unknown [2]. This can be explained by the shorter female urethra, greater urethral mobility and a relatively straight urethra that is well encased in the bony pelvis [3]. FUS though uncommon can cause life-impacting lower urinary tract symptoms. Patients with FUS often present with bothersome symptoms such as frequency, hesitancy, poor stream, urgency and dysuria that often culminate in UTI and urinary retention. Usually, local tissue fibrosis emanating from trauma, malignancy, radiation, iatrogenic injury and inflammatory diseases leads to

narrowing of the urethral lumen and malfunction [4,5]. Patients with HIV infection and Acquired Immunodeficiency Syndrome (AIDS) are predisposed to recurrent UTI [6]. Furthermore, the relatively short and straight female urethra increases susceptibility to UTI. Recurrent UTI precipitate periurethral inflammation and fibrosis leading to urethral narrowing. FUS contribute up to 18% of cases of bladder out

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Citation: Otele WH, Aswani JA & Ndiema RC. (2020) Recurrent Female Urethral Stricture Managed by Double Buccal Mucosa Graft Urethroplasty: Case Report. J Womens Health Safety Res, 4(2): 192-197.

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flow obstruction (BOO) in females [7]. Investigations are many and varied with no guidelines as to the most appropriate. The most common ones include urethrography, cystourethroscopy, uroflowmetry, urodynamic studies and ultrasonography to determine post-void residual (PVR) urine volume [8,9].

Novel- reconstructive surgical techniques have supplanted time-honored urethral dilation and CIC, both of which are not cost-effective [9,10]. Urinary Outcome of surgical reconstruction of FUS by local flaps and grafts, or remote grafts can be affected by complications such as urethral fistula and incontinence. Risk of urinary incontinence associated with female urethroplasty can be circumvented by in-lay procedures performed intraluminal unlike on-lay procedures performed outside. Although efficacy of surgical reconstruction is largely unknown, urethroplasty provides a higher mean success rate in the range of 80 to 94% compared to urethral dilation (less than 50%) in some series [10].

CASE PRESENTATION

We present the case of a 58-year-old woman (Initial M.W.G), para 4+0, Housewife. She was a known patient with HIV disease since early 2015 and was compliant on antiretroviral therapy with a normal viral load and CD4 count. She presented at the South B Hospital in Nairobi City in Kenya on 09.11.2018 with obstructive urinary symptoms. Previously she had experienced frequent episodes of UTI culminating in urethral strictures almost two years earlier and was being managed in another health facility with frequent urethral dilations but had declined CIC. She was in satisfactory general condition upon assessment. A flow rate of 7.5 ml/s on uroflowmetry, American Urological Association-Symptom Index (AUA-SI) of 25 and PVR urine volume of 118 ml against a pre-void volume of 315 ml with retention cystitis. Attempts at urethral catheter insertion or cystoscopy were futile. A VCUG showed a near obliterative distal urethral stricture, while Computerized axial Tomography scan of the pelvis showed a grossly distended urinary bladder with distal urethral narrowing but no other pathology (**Figures 1 and 2**).

Urodynamics could not be performed due to lack of supportive infrastructure. Renal ultrasound scan showed mild left hydronephrosis, but renal function tests were normal. A urine culture performed grew *E. Coli* sensitive to ciprofloxacin. This was treated with a full course of antibiotics and subsequent cultures confirmed eradication of the bacteria. Informed consent for BMG urethroplasty was solicited. Her regular physician concurred with decision for augmented urethroplasty. Upon consent, she was provided with Chlorohexidine oral gargles the day before surgery and on the morning of surgery (07/12/2018). At surgery, prophylactic antibiotics comprising Gentamicin 160 mg IV, Ciprofloxacin 200 mg

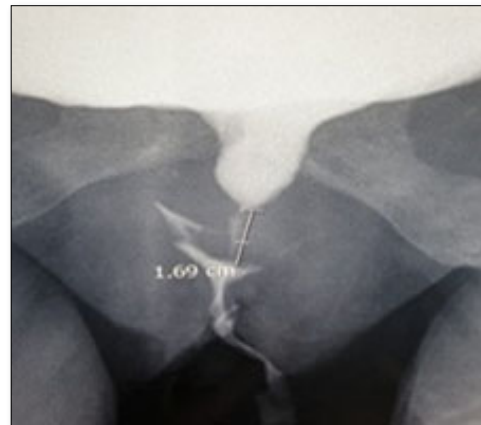


Figure 1. Voiding cystourethrogram (VCUG): Shows near obliterative distal urethra (1.69 cm long) and dilated upper ½ urethra plus grossly distended urinary bladder.

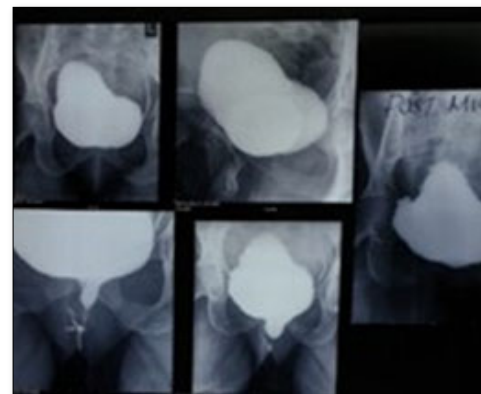


Figure 2. VCUG showing distal ½ urethral stricture and proximal dilation of the urethra, ballooning of bladder and diverticulation, left vesico-ureteric reflux and significant PVR urine.

IV and metronidazole 500 mg IV were administered. Patient was cleaned and draped aseptically. A semi-rigid ureteroscope 8F used for urethroscopy under spinal anesthesia confirmed distal near obliterative urethral stricture. Nylon 2/0 labial stay sutures were applied so as to facilitate exposure of the urethral lumen and vagina. With the aid of a dissecting forceps, incision was made at 12 o'clock with blade size 11 and extended proximally up-to and slightly beyond stricture site. A dense peri-meatal scar tissue on the right was excised (**Figure 4**). BMG was obtained from oral cavity a septically, defatted, divided into two and one quilted as in-lay graft on dorsal urethral wall and the other at the right peri-meatal area where scar tissue was excised (**Figures 5,6,7 and 8**). Donor site was primarily closed and packed with gauze for 4 hours. An indwelling 16 F two-way pure silicon urethral catheter was inserted and withdrawn after 21 days. Bactigras gauze was used for Peri-catheter wound dressing. A vaginal pack was left in-situ for

24 hours to prevent post-operative bleeding. Oral feeding was commenced after withdrawal of oral pack.



Figure 3. Stenotic urethral meatus.



Figure 4. Urethral meatus after excision of fibrous tissue from right urethra lip and dorsal stricturotomy.

PROCEDURE AND OUTCOME

The patient was discharged on third post-operative day and seen after 21 days for catheter removal. Thereafter, she was seen at 3 months, and every 6 months. During this time, the mean flow rate was 26 mls per second, Mean AUA-SI score 7 and insignificant PVR. Cystoscopy at 6 and 15 months showed a patent urethral lumen. Patient has not had significant obstructive symptoms to warrant further surgical interventions. Furthermore, she registers profound satisfaction by Patient Reported Outcome Measures.

DISCUSSION

Female urethral strictures are relatively uncommon and when they occur, significantly affect quality of life [11]. FUS are multifactorial in aetiology albeit rare in urological practice. In our case, frequent UTI in an immune compromised patient was the possible cause of



Figure 5. Buccal Mucosa Graft harvested.



Figure 6. Defating of BMG in preparation for grafting.

inflammation leading to the urethral stricture. HIV and AIDS predispose to recurrent UTI [6]. Surgical treatment of FUS involves either urethral dilation or reconstruction. Urethral dilation, the main stay treatment modality for a longtime has limited efficacy and is often associated with complications such as bleeding, extravasation and recurrent fibrosis and fistula formation [2,12]. Our patient had been on a regime of frequent urethral dilations for nearly 2 years without reward.

Osman et al in systematic analysis found a composite success rate of 47% at 43 months following dilation, worsening with subsequent dilatations [10]. Hoag and Chee in a review reported a 43% to 57% success rate after urethral dilation [9]. The higher success rate following urethral dilation may be attributed to concomitant use of CIC. Prior urethral dilation was a surrogate of failure in this group [13]. Popat and Zimmern reported a 43% durable resolution in their clients with outcome diminishing to 30% in repeat procedures [14]. Better success is achieved where duration of symptoms before presentation is shorter [14].



Figure 7. Harvesting Buccal mucosa graft from left cheek under local anaesthetic: 1% lidocaine/Adrenaline 1:100,000 solution.

Appropriate patient counseling on the unreliable efficacy of urethral dilation and need for auxiliary procedures such as CIC is essential. Furthermore, patients afflicted with FUS should be offered a suitable urethral reconstructive procedure rather than subjected to frequent urethral dilations [15]. Strictures in the mid-urethra may be amenable to endoscopic management with cold knife or laser ablation followed by catheterization with or without CIC. Urethrotomy combined with urethral dilation is associated with short term success but may culminate in urinary incontinence owing to sphincter damage. Meatoplasty is reserved for the distal urethral stricture [14,16].

A plethora of reconstructive procedures for FUS abound. They include vaginal flaps and grafts plus oral mucosal grafts. Vaginal flap or graft urethroplasty can be by ventral or dorsal approach, either producing comparative success rates. Vaginal Flap Urethroplasty can be combined with additional procedures such as dilation or pubo-vaginal slings or Martius flap as need may arise [9]. Both vaginal flaps and grafts depend on viable local tissues and would be unsuitable in the event of vaginal atrophy or scarification. Both techniques avoid morbidity associated with oral mucosa graft [9]. The overall success rate emanating from urethroplasty in females is estimated at 80% to 94% [10].

Women with FUS can also be managed by dorsal BMG; a safe, reliable and robust procedure. The dorsal BMG is technically feasible, well tolerated with a satisfactory outcome. Long-term follow-up is warranted with a larger patient series so as to critically interrogate patient outcomes [11]. BMG urethroplasty is usually a procedure of second resort after a failed urethral dilation. Although the procedure is not standardized in females as in males, ventral or dorsal graft approaches are feasible [11]. Risk of urinary incontinence can be mitigated by intraluminal in-lay approach to BMG. Where near obliterative strictures exist, double-face or more than one graft tissue can be applied as

in our present case (**Figure 9**). Joshi and Kulkarni reported a novel technique of double-face BMG for use in FUS in which two Buccal mucosa grafts were separately applied as dorsal on-lay and ventral in-lay [11]. Patient experienced normal unobstructed voiding at 6 months follow-up. Double face BMG is safe and efficacious when appropriately selected [11].



Figure 8. Neo-urethra with catheter in-situ, ready for grafting.

Goel et al. [17] in a single Centre experience performed dorsal on-lay BMG urethroplasty on 8 women with moderate to severe bothersome LUTS due to FUS and found a satisfactory outcome after a mean follow-up of 14.8 months. Patients experienced significant improvement in AUA-SI, uroflowmetry and PVR evaluation in the first 9 months post-operatively [17]. Hampson et al. [12] in a retrospective analysis of dorsal on-lay BMG in 39 patients with FUS performed by different surgeons found a success of 77% at 14 months mean follow-up period [12].

Agochukwu-Mmonu et al. in a systematic review confirmed efficacy of urethroplasty by graft or local flaps compared with urethral dilation [18]. Petrou et al in an evaluation of 11 women with FUS who underwent dorsal vaginal graft urethroplasty recommended the procedure as a first line option though no agreement or consensus exists on the choice of surgical treatment for FUS [19]. Tsivian et al. [20] performed Dorsal graft urethroplasty (Vaginal or Buccal) and established promising efficacy of the procedure though this series comprised only 3 patients.

No large comparative clinical trials have been conducted to establish the superiority of one technique over the other, leaving choice to be determined by the surgeon based on experience and preference [21]. Urethral stricture disease, though rare in females is a complex clinical condition that requires meticulous evaluation and judicious management [22]. Further research, elucidating disease etiology, pathophysiology, appropriate investigations, timely and



Figure 9. Grafted distal ½ of dorsal urethra and right lip of urethral meatus.

efficacious treatment protocols are necessary so as to mitigate morbidity associated with this condition.

Although the classic Double-face BMG urethroplasty technique was not applied in the foregoing case, double grafts were employed; one dorsally applied as in-lay from meatus to area beyond the strictured portion and another one on the right lip of the urethra where a dense fibrotic scar tissue was excised. Double female BMG urethroplasty is technically easy and safe to perform. Furthermore, it is cost-effective and definitely a better option to frequent urethral dilations.

ETHICS COMMITTEE APPROVAL

Ethics committee approval for this publication was sought from and granted by South B Hospital ethics committee (protocol number 001/2020).

AUTHOR CONTRIBUTIONS

Willy Otele- surgical procedure, concept design, supervision, manuscript proof reading Ndiema RC- Report analysis and interpretation. Aswani JA- literature search with critical review.

CONFLICT OF INTERESTS

The authors have no conflict of interest to declare.

FINANCIAL DISCLOSURE

Authors confirm that this study has received no financial support.

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