



Recurrent ventral curvature after corporoplasty with tunica vaginalis flap

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Keywords

Hypospadias; Island flaps; Pedicled flaps; Penis; Children

Abbreviations

TVF, tunica vaginalis flap; VC, ventral curvature; RVC, recurrent ventral curvature; IQR, interquartile range; SIS, small intestine submucosa; SF-1, Steroidogenic Factor-1; IRB, Institutional Review Board

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Summary

Introduction and objective

Optimal means to correct ventral curvature (VC) is debated. Our preferred technique for curvature greater than 45° is corporoplasty using tunica vaginalis flap (TVF). We describe our complications with TVF for ventral lengthening.

Methods

Forty-four boys who underwent ventral lengthening with a corporoplasty with TVF were identified in a prospective database for proximal hypospadias repair by a single surgeon from 2008 to 2021. Corporotomy was performed by incising the tunica albuginea of the corpora cavernosa transversely at the point of maximum curvature. Harvested TVF was tailored to the size of the corporotomy and anastomosed to the edges of the tunica albuginea and on laid to the corporal defect with the mesothelial side of the TVF abutting the erectile tissue.

Results

Median age at surgery was 1.0 years (IQR 0.72–1.82). Median follow-up time was 4.9 years (IQR 2.6–8.0). Thirteen patients (27%) were older than 10 years of age at last follow up (median 13.3, range 10–20). Twenty-two boys (50%) received pre-operative testosterone. The most common location of the meatus after degloving was penoscrotal (41%). Median VC after degloving was 90° (IQR 80–100).

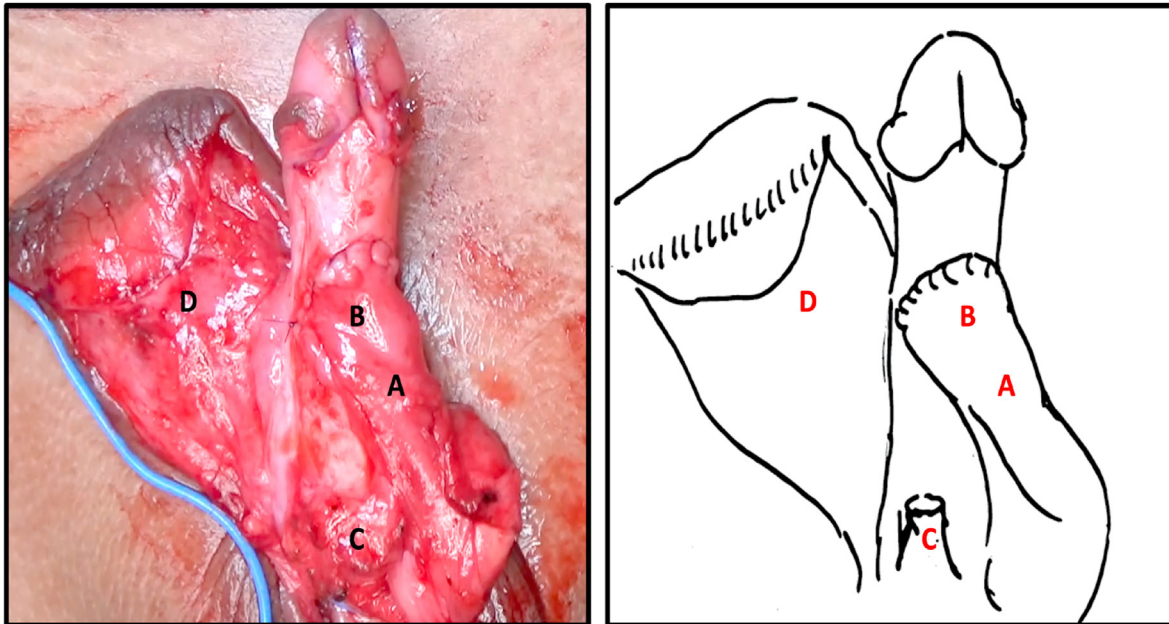
The urethral plate was transected in 43/44 (98%) of boys, improving median VC to 60° (IQR 40–60). After corporotomy, the median longitudinal distracted distance was 15 mm (IQR 12–17). Urethral reconstruction was most commonly achieved with the transverse island preputial flap technique or its modifications (39/44; 89%). Erections were reported in 42 boys (95%). None developed corporal diverticula, and two patients (4.5%) had ascended testis associated with TVF harvest. Seven percent of boys had recurrent ventral curvature (RVC; 3/44). Median RVC was 30° (IQR 30–45). One patient had RVC at the penoscrotal junction (not at site of prior corporoplasty) identified 11 years post operatively at age 15, and underwent dorsal plication. The other 2 patients were diagnosed less than 1 year post operatively. Both patients received testosterone due to small glans size, had double-face tubularized transverse island preputial flap as urethral and ventral skin coverage, and had endocrine and genetic consultation. Both had scarring of the preputial flap and of the corporoplasty. Scar excision and superficial transverse incisions on the tunica albuginea corrected RVC.

Conclusions

The five-year outcome of ventral penile lengthening using TVF for corporoplasty is favorable with 7% of boys with RVC, and 4.5% with ascended testes associated with TVF harvest. None developed corporal diverticula.

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Summary Figure Tunica vaginalis flap (TVF) as corporoplasty to correct ventral curvature. (A) Associated TVF pedicle is mobile and not on tension. (B) Terminal end of TVF sewn to corporoplasty. (C) Proximal end of transected urethral plate. (D) Preputial flap for coverage of ventrum of penis.

Introduction

Ventral curvature (VC) in hypospadias is thought to result from a growth disparity between the normal dorsal tissue of the corporal bodies and the attenuated ventral structures: abnormalities of the ventral skin or dartos causing shortening, tethering of the urethral plate, or disproportion of the corpora cavernosa [1]. Approximately 80% of patients with proximal hypospadias have associated VC [2]. Different techniques for ventral penile lengthening for correction of severe VC, typically defined as greater than 45°, are currently employed by pediatric urologists, most commonly deep transverse incision of the tunica albuginea or ventral corporotomy with grafting [3]. Various materials are available for corporoplasty, including homografts (dermis, tunica vaginalis as graft) and xenografts (small intestine submucosa (SIS), lyophilized bovine dura, bovine pericardium, cadaveric dermis) [4]. Our preferred technique for severe VC is corporoplasty using tunica vaginalis flap (TVF) [5].

Adolescents and adults who underwent distal and proximal hypospadias repair in childhood report that persistent VC significantly impacts their quality of life as compared to other domains, such as meatal position and shape [6]. We aim to report the incidence of recurrent ventral curvature (RVC), corporal diverticulum and ascended testis associated with TVF harvest in a consecutive cohort of patients who have undergone ventral penile lengthening for correction of VC with corporoplasty with TVF over 13 years. We also aim to describe common factors that may be associated with RVC in individuals who had RVC after TVF corporoplasty.

Methods

Boys who underwent ventral lengthening with a TVF corporoplasty were identified in a prospectively maintained database for proximal hypospadias repair by a single surgeon from 2008 to 2021 (IRB# 190105). Patients who had previously undergone hypospadias repair were included. Two to three doses of preoperative intramuscular 25 mg testosterone propionate were administered in the outpatient setting if the measured glans width was under 15 mm.

Data collection

Patient demographics, preoperative and operative clinical characteristics, and postoperative outcomes were collected. Follow-up phone calls were made to families of patient who were lost to follow up to discern reasons for attrition.

TVF corporoplasty operative technique and intraoperative data

The penis was degloved and an artificial erection test was performed. VC was measured with a goniometer. The corpus spongiosum was dissected off the corpora cavernosa. The urethral plate was divided at point of maximum curvature. The distance between the ends of the distracted transected urethral plate was recorded with the penile shaft under manual traction to simulate corpora cavernosa with no VC nor dorsal curvature. A repeat artificial erection was performed to measure VC again after transection of the

urethra, and to mark the new point of maximum curvature in preparation for corporotomy should VC persist.

We have previously described our technique for TVF harvest and use in corporoplasty [5,7]. To minimize tourniquet time on the penile shaft, the TVF was harvested prior to performing the corporotomy. A transverse, broad-based incision was made in the tunica vaginalis anteriorly at the inferior pole of the testis. The tunica vaginalis incision was carried proximally on either side of the spermatic cord towards the external inguinal ring to maximize the mobility of the pedicle.

Subsequently, a tourniquet was placed on the penile shaft. A transverse incision was made on the tunica albuginea at the point of maximum curvature from the 3 to 9 o'clock position. The tunica albuginea was meticulously elevated off the vascular erectile tissue proximally and distally. The maximal longitudinal distance between the proximal and distal edges of the corpora cavernosa was measured with the penile shaft under manual traction." The TVF was rotated so that the mesothelial layer (visceral layer) of the tunica albuginea abutted the vascular erectile tissue (Fig. 1A). Using a double-armed 6-0 polydioxanone suture, the "apex" of the TVF was secured (Fig. 1B). Unlike free grafts, where typically an additional 25% of surface area of the graft is allocated to account for contraction during healing [8], the TVF was tailored to the surface area of the corporotomy (Fig. 1C and D). The anastomosis was completed in a running locking manner circumferentially (Fig. 1E). In all operations the TVF was used as a stand-alone coverage without other materials or grafts present (Fig. 2).

No artificial erection test was performed after corporoplasty was completed. The remaining ventral coverage was performed at the surgeon's judgment, but typically involved a variation of the transverse island preputial flap when prepuce was available.

Postoperative outcomes

The primary outcome in our study was RVC of any degree that occurred anytime following corporoplasty with TVF. If

the patient returned to the operating room for further penile procedures, such as a second stage hypospadias repair or urethroplasty, the degree of curvature elicited from the erection test was recorded. All VC was objectively measured with a goniometer. If the patient did not have further procedures, such as those undergoing a single-stage double-face onlay-tube-onlay tubularized transverse preputial island flap, the presence of straight erections or curvature were determined by parental history and/or photographic documentation.

Secondary outcomes were corporal diverticulum formation and ascended testis associated with TVF corporoplasty. Corporal diverticulum was defined as any outpouching of the tunica albuginea that did not follow the contour of the corpora cavernosa with erections. This was primarily assessed on artificial erection test if the patient underwent subsequent penile surgery; if unavailable, we relied on parent history and photographs. Ascended testis associated with TVF were defined as the need for subsequent orchopexy postoperatively on the same side that the TVF was harvested.

Statistical analysis

Descriptive statistics were summarized using number (percentage) for categorical variables and median (interquartile range) for continuous variables. Data was analyzed using SAS Statistical Software version 9.4 (SAS Institute, Cary, NC, USA).

Results

Pre-operative characteristics

Among the 906 boys identified in our database, 194 had proximal hypospadias. Forty-four boys who underwent ventral lengthening with a TVF corporoplasty were identified. Patient demographics and clinical characteristics are listed in Table 1. Median age at surgery was 1.0 years (interquartile range (IQR) 0.72–1.82), and median follow-up time was 4.9 years (IQR 2.6–8.0). Thirteen patients

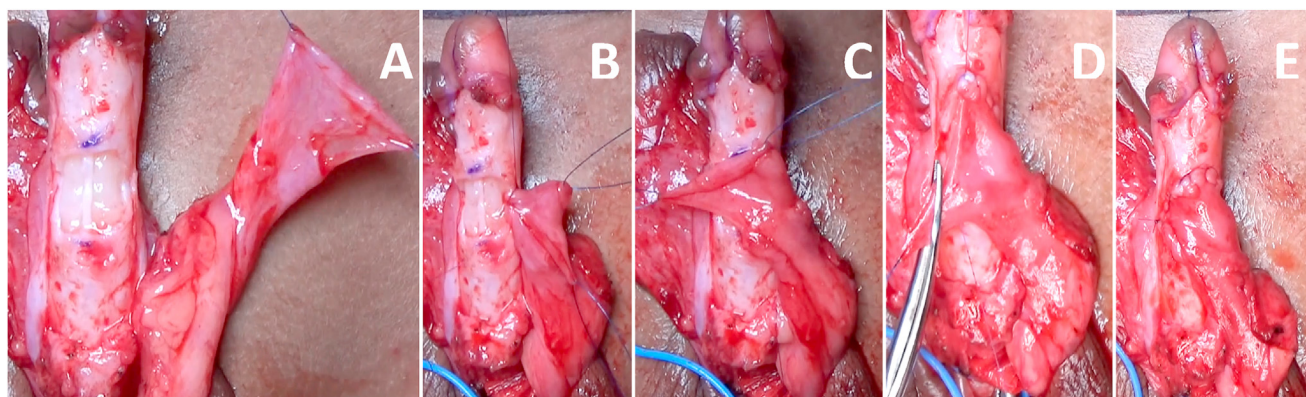


Fig. 1 Tunica vaginalis as pedicled flap in corporoplasty for ventral curvature repair. (A) The TVF is then rotated so that the mesothelial layer (visceral layer) of the tunica albuginea abutted the vascular erectile tissue. (B) Using a double-armed 6-0 polydioxanone suture, the "apex" of the TVF is secured. (C) The flap is laid over the corporotomy to gauge its fit. (D) The tunica vaginalis flap is tailored to the surface area of the corporotomy. Unlike free grafts, no substantial additional surface area is needed to account for subsequent contracture. (E) The anastomosis is completed in a running locking manner circumferentially.

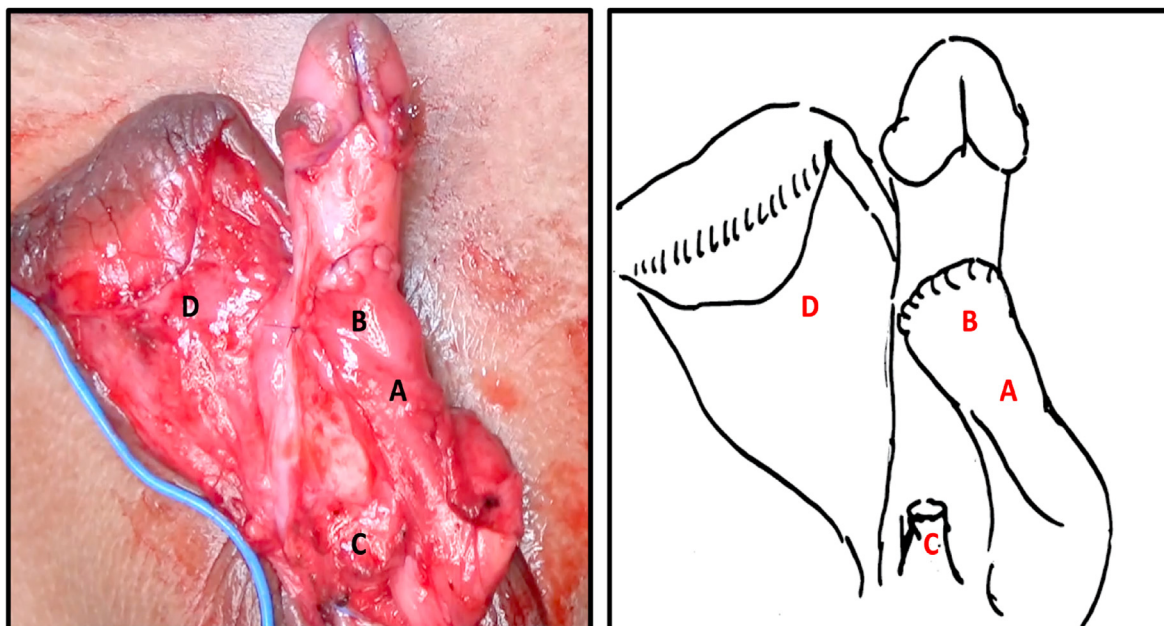


Fig. 2 Tunica vaginalis flap (TVF) as corporoplasty to correct ventral curvature. (A) Associated TVF pedicle is mobile and not on tension. (B) Terminal end of TVF sewn to corporoplasty. (C) Proximal end of transected urethral plate. (D) Preputial flap for coverage of ventrum of penis.

(27%) were older than 10 years of age at last follow up (median 13.3, range 10–20). Twenty-two boys (50%) received preoperative testosterone prior to surgery.

The most common concurrent condition was the presence of undescended testes (11%). Four boys (7%) had prior hypospadias operations. All prior procedures (4/4, 100%) were proximal hypospadias performed without ventral penile lengthening for correction of VC. Median preoperative glans width was 15 mm (IQR 12–17 mm). Preoperative meatal location was most commonly present in the penoscrotal junction (41%), followed by a perineal location (18%) and scrotal location (14%).

Operative characteristics

Operative characteristics are detailed in Table 2. The most common location of the meatus after degloving remained penoscrotal (41%), followed by scrotal (27%) and perineal (21%). Median VC prior to degloving was 90° (IQR 80–100°), without significant improvement after degloving (median VC 90°, IQR 50–100°). The urethral plate was transected in 43/44 patients (98%), with improvement of median VC to 60° after urethral transection (IQR 40–60°). The median distracted distance between the distal and proximal edges of the transected urethral plate was 40 mm (IQR 30–45 mm). After corporotomy at the point of maximal curvature on the ventral surface, the median longitudinal distracted distance between the edges of the tunica albuginea after release of the underlying erectile tissue was 15 mm (IQR 12–17 mm). Ventral surface skin coverage or urethral reconstruction, at the discretion of the surgeon, was most commonly achieved with a double-face onlay-tube-onlay transverse preputial island flap (57%), followed by a first stage hypospadias repair with a transverse island preputial flap (32%).

Outcomes

The presence of erections was reported in 42 boys (95%), with 17/42 (40%) of patient having erection tests performed in subsequent penile surgeries for verification, 3/42 (7.1%) having clinical photographs and clinical history, and 22/42 (52%) with parental clinical history only as verification.” In the remaining two individuals (5%) where erections could not be verified, the families could not be reached due to relocation out of the country and a disconnected line at the last known phone number. Three boys had RVC after TVF corporoplasty (7%). Median RVC was 30° (IQR 30–45). One patient had RVC of 45° occur at age 15 years, identified 11 years after the initial TVF corporoplasty and first stage hypospadias repair with preputial island flap. His reoperation for correction of RVC was a dorsal Nesbitt plication, with follow up demonstrating successful correction. The point of maximal curvature was noted to be at the penoscrotal junction, 5 cm proximal to the previous site of operation of the TVF corporoplasty. He had no other medical comorbidities.

The remaining two patients with RVC were diagnosed less than one year post operatively. Both received testosterone due to small glans size in the initial operation and had double-face tubularized transverse island preputial flap as urethral and ventral skin coverage. On endocrine workup for disorders of sexual differentiation, one had a gene mutation that codes for the Steroidogenic Factor-1 (SF-1 protein), which is central to the regulation of steroid synthesizing genes. Another had genetic consultation for the constellation of hemivertebrae, facial hemangioma, short stature, and hypospadias, with no conclusive diagnosis. In reoperation for correction of RVC, both patients had scarring of the preputial flap and of the corporoplasty. Scar

Table 1 Patient baseline demographics and clinical characteristics.

Demographic Data	
Race/ethnicity, n (%)	
Hispanic, any race	14 (32)
Non-Hispanic white	17 (39)
Asian	8 (18)
African American	0 (0)
Unknown	5 (11)
Preoperative	
Clinical Characteristics	
Median age at surgery, years (IQR)	1.0 (0.72–1.82)
Median age at last follow-up, years (IQR)	7.4 (3.7–9.9)
Median duration of follow-up, years (IQR)	4.9 (2.6–8.0)
Received preoperative testosterone, n (%)	22 (50%)
Concurrent conditions ^a	
Undescended testis	5 (11)
Disorders of sexual differentiation	4 (9.0)
Mixed gonadal dysgenesis	1 (2.0)
Hemophagocytic lymphohistiocytosis	1 (2.0)
Cardiac conditions	1 (2.0)
Previous hypospadias operation, n (%)	4 (9.0)
Median glans width, mm (IQR)	15 (12–17)
Meatal location prior to intervention, n (%)	
Distal	5 (11) (2 penoscrotal prior to 1st surgery)
Mid-shaft	3 (7.0) (1 penoscrotal prior to 1st surgery)
Penoscrotal	18 (41)
Scrotal	6 (14)
Perineal	8 (18)
Proximal (unspecified)	4 (9.0)

^a Patients may have multiple conditions.

excision and deep transverse incisions on the tunica albuginea corrected RVC with verification of straight erections in subsequent second stage urethroplasties.

None of the patients developed corporal diverticula at time of last follow up. Two boys had subsequent orchiopexy on the same side that the TVF was harvested from (4.5%). The three most common secondary complications in our cohort were urethrocutaneous fistula formation (23%), repair breakdown (18%) and urethral stricture formation (16%) (Table 3). The total complication rate in this cohort is 55%.

Discussion

Our current series represents the largest cohort of patients who underwent TVF for corporoplasty in ventral penile

Table 2 Operative characteristics of patients who underwent TVF corporoplasty for correction of ventral curvature.

Operative Characteristics	
Meatal location after degloving, n (%)	
Mid-shaft	1 (2.0)
Penoscrotal	18 (41)
Scrotal	12 (27)
Perineal	9 (21)
Proximal (unspecified)	4 (9.0)
Median degree of curvature prior to degloving, degrees (IQR)	90 (80–100)
Median degree of curvature after degloving, degrees (IQR)	90 (50–100)
Median cut urethral plate distance distracted, mm (IQR)	40 (30–45)
Urethral plate not cut, n (%)	1 (2.0)
Median degree of curvature after transection of urethral plate, degrees (IQR)	60 (40–60)
Urethral plate not cut, n (%)	1 (2.0)
Median distracted distance of tunica albuginea of corpora cavernosa after corporotomy, mm (IQR)	15 (12–17)
Subsequent operation of coverage over TVF, n (%)	
First stage hypospadias repair with transverse preputial island flap	14 (32)
Double-face onlay-tube-onlay tubularized transverse preputial island flap	25 (57)
Other (eg. Z-plasty, first stage buccal mucosal, Byar's flap)	5 (11)

Abbreviations: TVF = tunica vaginalis flap; IQR = interquartile range.

lengthening in the literature [5,9–11]. TV demonstrates good tensile properties, is highly vascular, does not contain hair follicles, and is located close to the penis [12]. It may be challenging to harvest in the setting of concomitant bilateral undescended testes. The purported advantage of using TV as a flap for corporoplasty is twofold. First, as compared to autologous dermal grafts, TVF minimizes donor site morbidity. Second, as compared to xenografts or homografts, the vascular supply provided by the TVF pedicle may theoretically minimize contracture. This was demonstrated in a comparative rabbit model examining histological sections of TV use as a flap versus its use as a graft in corporoplasty. The authors found no contractures in the TVF group, whereas the TV graft group had progressively worsening contractures with increased healing time, with 42% contracture of the graft at 12-week autopsy specimen [12]. A disadvantage of using TVF for corporoplasty as compared to grafts is the inability to perform a repeat artificial erection test once the TVF is sewn in place. Despite this operative limitation, 93% of our patients had straight erections and none developed corporal diverticula.

Table 3 Recurrent ventral curvature, corporal diverticulum formation and ascended testis in patients who underwent corporoplasty with tunica vaginalis flap.

Postoperative outcomes and complications	
Having erections, n (%)	
Yes	42 (95)
No	
Missing	2 (5.0)
Recurrent ventral curvature, n (%)	
Yes	3 (7.0)
No	41 (93)
If recurrent ventral curvature,	
Median degree of curvature, degree (IQR)	30 (30–45)
Re-operation for ventral curvature, n (%)	3/3 (100)
Corporal diverticulum formation, n (%)	0
Subsequent orchiopexy on same side that TVF was harvested, n (%)	2 (4.5)
Additional complications, n (%) ^a	
None	20 (45)
Breakdown	8 (18)
Fistula	10 (23)
Stricture	7 (16)
Graft scarring	2 (5.0)
Urethral diverticulum	1 (2.0)
Scrotal abscess	1 (2.0)
Meatal stenosis	1 (2.0)

Abbreviations: TVF = tunica vaginalis flap; IQR = interquartile range.

^a Some patients had multiple complications.

We have previously reported on 23 patients who underwent TVF alone for corporoplasty, one of whom had RVC due to scrotal skin tethering with a RVC rate of 4.3% [5]. In the current larger series of patients, we demonstrate a slightly worse RVC of 7% with a median follow up of 4.9 years, which is reasonably favorable when compared with other methods of correction of VC. With a mean follow up of 44 months, Snodgrass and Bush recently reported on their aggregate RVC rate of 26%–36% when performing a “proximal tubularized incised plate” technique [13]. Among their cohort, 23% (7/31) developed RVC after undergoing dorsal plication alone, 25% RVC rate (5/20) in those who had dorsal plication in addition to other maneuvers, and 43% (3/7) RVC rate in those who had urethral plate elevation with or without corporotomies without dorsal plication.

In comparing RVC to other grafts used for corporoplasty, most studies are plagued by short term follow up and a small number of patients [8,14]. With a mean follow up of 55 months in 153 patients who underwent corporoplasty with 1-ply SIS, Suarez et al. reported an RVC rate of 6.5% [15], which is comparable to the TVF corporoplasty rate in our series of 7%.

The importance of a longer duration of follow up in hypospadias repair, specifically proximal hypospadias, has increasingly received attention in the past decades [16,17]. Although up to 50% of postoperative complications are identified in the first post-operative year, longer follow up has yielded higher rates of complications [17,18]. In a series

of 22 patients who presented with chordee 10 years after their initial hypospadias operation, Vandersteen and Husmann noted that median age at presentation was 21 years [19]. In our series one patient had RVC occur during puberty, 11 years after his initial operation with TVF corporoplasty. The point of maximal RVC during reoperation was 5 cm proximal to the original site of corporotomy and corporoplasty, suggesting disproportionate corporal growth during accelerated penile growth at puberty as a probable cause. We aim to extend follow up in this cohort of patients.

In both patients who had RVC related to TVF scarring and preputial flap scarring, both had phenotypic manifestations that prompted endocrine or genetic workup. On endocrine evaluation, one patient was found to have a gene mutation affecting SF-1 expression. SF-1 is a nuclear factor that has a central role in gonadal and adrenal expression, and mutations in its encoding gene (NR5A1) leads to down-regulated expression of SF-1, which has been implicated in the embryologic development of hypospadias [20,21]. The remaining patient was referred for genetic consultation for a constellation of symptoms. In our retrospective cohort study without a comparison group nor complete genomic data on patients who did not develop RVC, it is challenging to draw causal inferences. The role of androgens in wound-healing of hypospadias is inconclusive. Some studies cite parenteral testosterone or topical dihydrotestosterone as yielding fewer complications such as wound dehiscence, urethrocutaneous fistula, and better cosmetic outcomes due to lower rates of scar formation [22,23]. In theory, androgen administration prior to hypospadias repair enlarges the penis and the glans, hence making repair less difficult, and decrease the possibility of complications. However, in a prospective study where children with distal and proximal hypospadias were randomized to receipt of androgen treatment, Kaya et al. found that scar formation after hypospadias repair was significantly lower in the group that received androgen therapy (2 of 37 patients, 5%) as compared to the control group (16 of 38 patients, 42%; $p < 0.05$) [23], raising the possibility for future investigation into exploration of the role of androgens in wound-healing.

Two boys had subsequent orchiopexy on the same side that the TVF was harvested from, suggesting an ascended testis rate of 4.5%. Although our sample size is small, we propose two theories as to its association with TVF corporoplasty. First, insufficient mobilization of the TVF and its associated pedicle can lead to tethering of the spermatic cord as the child grows. Second, that scarring of the TVF pedicle or associated components leads to tethering of the testis.

Our study has several limitations. First, about 70% of our cohort were not yet past age 10 years, with probably fewer when stratified by Tanner stage who had reached pubertal age. Pubertal penile growth can magnify issues caused by corporal disproportion and manifest delayed onset RVC. Second, we acknowledge that our study does not account for the quality of erections and its suitability for penetrative intercourse in the future. In the future, we aim to administer the Modified Erection Hardness Score in post-pubertal boys who have undergone hypospadias repair. Third, although we attempted to mitigate the issue of

differential loss to follow up (i.e., those who did not follow up had worse outcomes) through contacting families, there were still some patients who were unable to be reached. Fourth, some patients had only erections subjectively reported on the basis of parental history, which is less reliable than clinical photographs or artificial erection tests.

Conclusions

In a cohort of 44 patients who underwent ventral penile lengthening using TVF for corporoplasty, 93% had straight erections on five-year follow up. The RVC rate of 7% in the intermediate term is favorable. None developed corporal diverticula. 4.5% of patients had ascended testes associated with TVF harvest.

Ethical approval

Ethical approval was not required. The study was approved by the Institutional Review Board IRB# 190105.

Funding

None.

Conflicts of interest

The authors declare no conflict of interest.

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