



A meta-analysis comparing dorsal plication and ventral lengthening for chordee correction during primary proximal hypospadias repair

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Abstract

Purpose Recurrent chordee (RC) is an important complication of proximal hypospadias repair. In this meta-analysis we compared RC incidence following dorsal plication (DP) versus ventral lengthening (VL).

Methods We searched the databases to identify all papers between 2001 and 2021 pertaining to proximal hypospadias and recurrent chordee. Duplicate publications, review articles and incomplete articles were excluded. Meta-analysis of heterogeneity was reported with I^2 statistics. The pooled outcomes were compared to Chi square/Fishers exact test.

Results A total of 17 articles were included covering 582 patients. The I^2 statistics for prevalence of RC among different publications showed no heterogeneity for DP ($I^2=0\%$) and low heterogeneity for VL ($I^2=26\%$). RC was noticed in 31/122 (25.4%; 95% CI 18%–33%) among patients who had DP alone while it was significantly lower, 24/460 (5.3%; 95% CI 4%–8%) when VL was used ($p=0.0001$). When compared to DP, all VL techniques had significantly lower incidence of RC. Among the VL techniques lowest incidence of RC was found for ventral corporotomies (4%) followed by small-intestinal- submucosa (SIS 4.2%) and tunica vaginalis flap (TVF)/free graft-TVFG (5%). Among the VL subtypes: the proportion of RC with use of TVF (4/70, 5.7%) and TVFG (3/69, 4.3%) for corporoplasty was comparable ($p=1$); single-layer SIS was associated with significantly less RC (1/90, 1.1%) than 4-layer SIS (5/51, 9.8%; $p=0.02$).

Conclusion For correction of severe ventral chordee during primary proximal hypospadias repair, dorsal plication carries a higher risk of recurrence compared to ventral lengthening procedures.

Keywords Proximal hypospadias · Chordee · Recurrence · Dorsal plication · Ventral lengthening

Background

Adequate correction of ventral chordee (VC) is essential during primary repair of proximal hypospadias. While several surgeons omit the important step of artificial erection test to objectively assess the chordee and its correction during primary repair, naked eye estimation is often prone to mistakes [1]. Late occurrence of recurrent chordee (RC) during adolescence has been reported by several authors [2, 3] causing cosmetic disfigurement and painful erections. Inadequate

and inappropriate correction of VC during primary procedure is an important cause of RC [4]. Vandersteen and Husmann [5] felt that a successful artificial erection induced at hypospadias surgery may not prevent RC which may occur late due to skin tethering, ventral fibrosis or corporeal disproportion and extensive urethral fibrosis.

There are multiple methods to correct VC during primary proximal hypospadias repair. While dorsal plication (DP) techniques may be applied for VC less than 30 degree [2], the urge to correct proximal hypospadias in one stage may make the surgeon underestimate the degree of chordee and choose DP and urethral plate preservation techniques [6] even in severe cases. Several ventral lengthening (VL) methods like ventral fairy cuts (corporotomy), ventral corporal excision/grafting (corporoplasty) have been used with or without urethral plate preservation [7, 8] for correction of VC more than 30 degree. In this systematic review and meta-analysis we have compared the contemporary outcomes of

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VC correction techniques during primary proximal hypospadias repair in preventing RC.

Methods

Search strategy

A systematic literature review of publications in English of the following electronic databases was conducted: Cochrane Database, PUBMED, MEDLINE and EMBASE. The following keywords were used: (proximal) AND (hypospadias) AND (chordee OR curvature) AND (recurrence OR complications). The publication date range for studies was from January 2001 to September 2021. Two researchers independently screened articles for the review.

Inclusion and exclusion criteria

We defined study eligibility using the population, intervention, comparator, outcome, and study design approach (PICOS). Population (P) was children with proximal hypospadias which included proximal penile, penoscrotal or perineal opening after degloving of the penis (hypospadias with severe chordee, in whom VC persisted after complete degloving/ventral skin detethering). Articles published before twenty years from present were not included as we were only interested in the contemporary outcomes in the new millennium.

Interventions (I) were chordee correction techniques along with proximal hypospadias. Only articles that mentioned chordee identification and correction technique were included. They included DP technique alone, which may be a midline plication (Baskin) or lateral plication (Nesbit) along with single stage repair; or VL techniques which may be ventral corporotomies, ventral fairy cuts, deep transverse incision of tunica albugenia (DTITA), ventral corporal excisions followed by bridging of the corporal defect (corporoplasty) with tunica vaginalis flap (TVF), tunica vaginalis free graft (TVGF), dermal/dural graft (DF), or small-intestinal submucosa (SIS). Papers not providing clear details of type of hypospadias, type of urethroplasty or VC correction technique were excluded [3, 6–8]. Isolated case reports, small case series (<5), those which included distal hypospadias also [9, 10], and review articles without original data were excluded.

Comparator I was between DP and VL techniques of chordee correction. Outcomes (O) analyzed was RC during follow-up after primary proximal hypospadias repair. RC was either diagnosed by the surgeon during outpatient follow-up/during the second procedure or complained by the patient/parent during the follow-up. Papers which did not provide the follow-up duration or incidence of RC in the

complication were excluded. Papers focusing on RC management alone could not be used in the meta-analysis on RC incidence without a denominator.

Study appraisal and synthesis

Study design (S) followed the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) criteria [11]. Study selection was performed through two levels of screening. In the first level, abstracts were reviewed for the inclusion and exclusion criteria. In the second level screening, all articles filtered through the first level were read in their entirety and the same inclusion and exclusion criteria applied. Figure 1 describes the methodology followed in this systematic review and meta-analysis.

Risk of bias analysis

Risk of bias (ROB) analysis was performed for each article by two authors separately and vetted using robvis tool [12] before inclusion in the meta-analysis. Six factors were assessed. Clarity of study population (pediatric), type of hypospadias (proximal), details on incidence of RC and duration of follow-up were expected to be essential and have low ROB. Urethroplasty technique and VC correction technique were considered important and was expected to have low or medium ROB. Following ROB analysis four papers [13–16] which combined both VL and DP techniques were considered to have high overall ROB and were excluded.

Data analysis

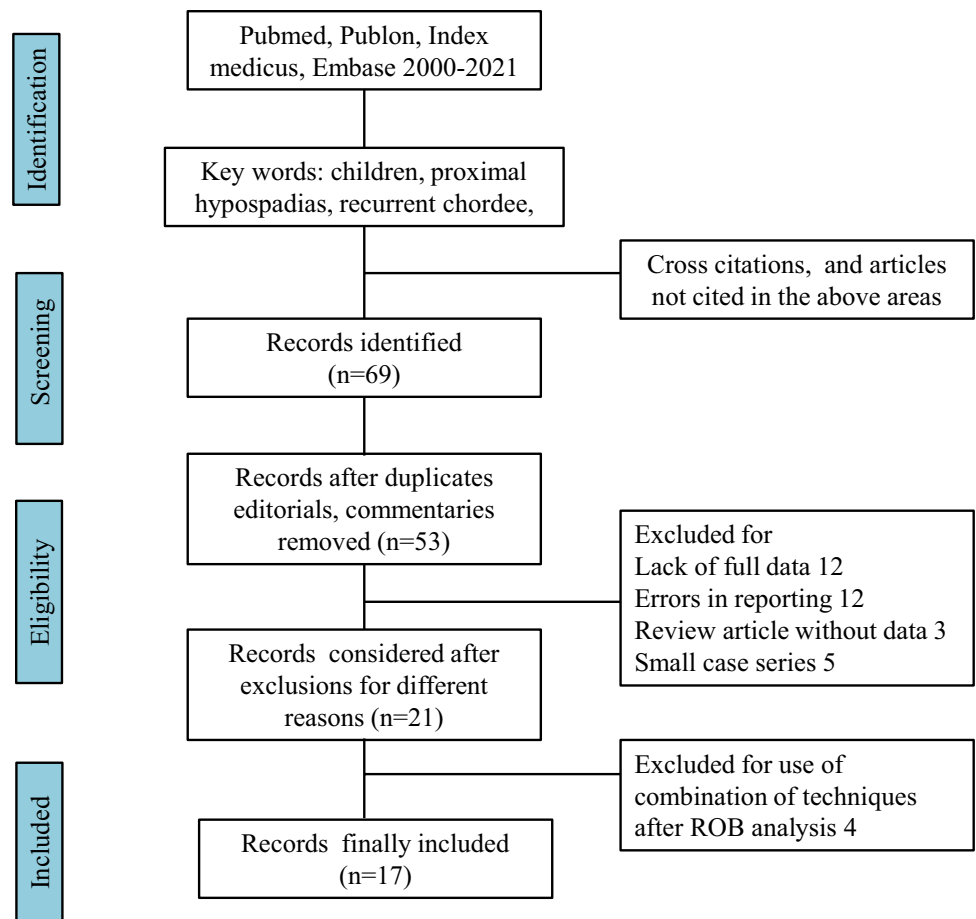
Meta-analysis of proportions was carried out using MetaXL 5.3 (Epigear International © 2010–2016). Heterogeneity was reported with I^2 statistics, with 0–25% not important, 25–50% moderate, 50–75% substantial, and 75–100% considerable. Forest plot was constructed to compare the pooled prevalence of complications in different VC correction methods used to treat proximal hypospadias. Statistical analysis comparing proportions was performed with Chi square test or Fisher's test when the values were small. The difference was considered statistically significant if the p value was <0.05.

Results

Study selection

Figure 1 shows the study selection process. Among 69 articles identified on screening, 16 articles were excluded as they were duplicates or editorials or commentaries without data. Due to inadequate data on primary repair/RC incidence

Fig. 1 PRISMA flow chart depicting study selection and inclusion



12 articles were excluded. Further 12 articles with errors, 3 review articles and 5 small case series (less than five patients) were excluded. After leaving further 4 articles due to overlapping VC correction techniques (ROB), a total of 17 articles [17–33] were included in this meta-analysis. All the studies reported children who typically underwent primary hypospadias repair between 9–18 months of age. All articles mentioned intra operative artificial erection test to assess VC & RC. Four articles [34–37] reporting only late RC and its management were excluded but were discussed separately as they did provide some useful information. The authors were from across the globe from many countries, as shown by the geographical origin of the included articles: USA, UK, Canada, Egypt, Japan and Turkey.

Risk of bias (ROB) analysis

Figure 2 shows the traffic light plot representing ROB of each article considered in the study. Four papers had high ROB as they described both DP & VL and were not included in the meta-analysis. In two studies with medium ROB [17, 20] only details regarding DP were considered;

as the remaining patients had a combination of procedures to correct chordee. All the remaining included articles had an acceptable overall ROB.

Meta-analysis

Figure 3 shows the forest plot for all included publications. The I^2 statistics for prevalence of RC among different publications showed no heterogeneity for DP ($I^2 = 0\%$) and low heterogeneity for VL ($I^2 = 26\%$). Publication bias studied with DOI plot showed Luis Furuya-Kanamori (LFK) index of 0.26 for DP and 0.69 for VL suggesting no asymmetry among papers making a good case for pooled meta-analysis with fixed effects model.

The pooled data from 17 articles covered a total of 582 patients with proximal hypospadias and severe VC. Table 1 describes the type of urethroplasty, primary VC correction technique and incidence of RC. The follow-up duration ranged from 12–120 months. Among all the included patients RC was reported in 55/582 patients which amounted to 9.4% (95% CI 7%–12%).

	Risk of bias						Overall
	D1	D2	D3	D4	D5	D6	
Snodgrass 2021	+	+	+	-	+	+	-
Badawy 2020	+	+	+	-	+	+	+
Al Adl 2020	+	+	+	X	+	+	X
Snodgrass 2017	+	+	+	+	+	+	+
Pippisalle 2016	+	+	-	-	+	+	-
Tiryaki 2016	+	+	+	X	+	+	X
Springer 2013	+	+	+	X	+	+	X
Rynja 2012	+	+	+	X	+	+	X
Miguel 2011	+	+	-	+	+	+	+
Hayn 2019	+	+	+	+	+	+	+
Hayashi 2010	+	+	-	+	+	+	+
Leslie 2008	+	+	+	+	+	+	+
Braga 2008	+	+	-	+	+	+	+
Badawy 2008	+	+	+	+	+	+	+
Elmore 2007	+	+	+	+	+	+	+
Braga 2007	+	+	+	+	+	+	+
Kajbafzadeh 2007	+	+	-	+	+	+	+
Weiser 2003	+	+	+	+	+	+	+
Soergel 2003	+	+	+	+	+	+	+
Ritchev 2003	+	+	+	+	+	+	+
Gershbaum 2002	+	+	+	+	+	+	+

D1: Study population clarity
 D2: Type of hypospadias clarity
 D3: Hypospadias repair technique uniform
 D4: Chordee Correction type uniform
 D5: Recurrent chordee numbers reported
 D6: Follow up data complete

Judgement
 X High
 - Unclear
 + Low

◀**Fig. 2** Risk of bias (ROB) analysis. Four articles with high overall ROB (two techniques VL and DP combined) were not included in the meta-analysis. Among two articles with medium ROB (VL or DP but described separately) data describing either one technique clearly was derived

Comparison of RC incidence between DP and VL techniques

Table 2 summarizes the pooled outcomes of VC correction techniques. Among patients who had DP alone as primary technique 31/122 had RC with an incidence of 25.4% (95% CI 18%–33%). Among patients who had VL as primary technique 24/460 had RC with an incidence of 5.3% (95% CI 4%–8%). VL techniques were associated with a significantly lower incidence of RC ($p=0.0001$) compared to DP.

Comparison of different VL technique

Among the VL techniques lowest incidence of RC (Table 2) was found for ventral corporotomies (4%) followed by SIS (4.2%) and TVF/TVFG (5%). Dermal/dural graft had 10.9% incidence of RC. When compared to DP, all VL techniques had significantly lower incidence of RC ($p<0.00001$ for ventral corporotomy/TVF/TVFG/SIS; $p=0.028$ for dermal/dural graft) but there was no significant difference within VL techniques.

Comparison of VL subtypes

The proportion of RC with use of TVF (4/70, 5.7%) and TVFG (3/69, 4.3%) for corporoplasty was comparable with no statistically significant difference ($p=1$). Although overall RC with SIS was comparable with other VL techniques, within SIS subgroup: single-layer SIS was associated with significantly less RC (1/90, 1.1%) than 4-layer SIS (5/51, 9.8%; $p=0.02$).

Etiology and management of RC

Although RC management is not the aim of this meta-analysis, we felt the four excluded publications [34–37] on RC management may be worth reporting separately (Table 3). Among 170 patients with RC, 100 (59%) were following DP and 33 (19%) were following VL. In 74 (43%) the type of primary VC correction technique was unclear. Skin scarring and ventral subcutaneous fibrosis were reported to be cause of RC in 73 (43%) of patients. In 54 (31%) of patients skin detethering ± DP was performed to correct RC. Corporal disproportion was reported in 42/56 (75%) in one study [35] which recommended staged repair with ventral corporotomies to treat RC.

Discussion

Ventral chordee is an important aspect to be addressed during proximal hypospadias repair. Skin detethering alone released VC in 19%, while in 31% it persisted but less than 30 degree and in 50% it was more than 30 degree [1]. The incidence of RC varies from 0 – 30% depending on the length of follow-up and the method of assessment [17, 25]. Several authors [2, 38, 39] have felt that poor intra operative assessment of VC and post-operative documentation of RC are responsible for under reporting of this particularly devastating complication. It is easy to underestimate lesser degrees of bending, as up to 30 degree of curvature can be easily mistaken to be inconspicuous on visual inspection [35]. While failure to perform artificial erection or objectively assess chordee using a goniometer during primary repair is an important reason to miss VC, inadequate/inappropriate correction of VC is an equally important cause. A survey [40] of pediatric urologists regarding VC correction reported that DP was their preferred management in those up to 40 degree of chordee.

DP was initially described by Nesbit on lateral aspects of corpora and was further modified by Baskin to include only midline 12 O' clock plication to safe guard nerves. In this meta-analysis we found that when DP was used to correct chordee in primary proximal hypospadias, it was associated with 25% incidence of RC and this was five times more than that found with VL procedures (5.3%). Braga[25] in a multivariate analysis also reported high odds (OR 4.5) of developing RC following DP compared to VL.

There are several VL procedures to correct severe chordee associated with proximal hypospadias. VL procedures like fairy cuts on corpora (corporotomy) and wide excision of corpora with grafting (corporoplasty) have gained popularity over the years for severe VC [18–20, 28]. In this meta-analysis we found that all VL procedures had significantly reduced incidence of RC. Among the various VL procedures, transverse ventral corporotomies had the lowest incidence (4%) of recurrent chordee while dermal/dural grafts had the highest (10.9%). While using tunica vaginalis for corporoplasty, it may be used as a vascularised flap (TVF) or as a free graft (TVFG). We found no difference in outcome between either TVF or TVFG, thereby supporting the use of the simpler TVFG which is easier to harvest and carries less risk of complications like twist, testicular ascent and acquired chordee/torsion known with TVF. Similarly, we also found that 4-layer SIS had higher risk of RC, probably due to fibrosis and graft contracture [24].

Despite adequate correction (confirmed by repeating artificial erection test) of VC during primary repair, RC is known to develop around adolescence in a proportion of patients [33, 34, 36]. While most papers included in this

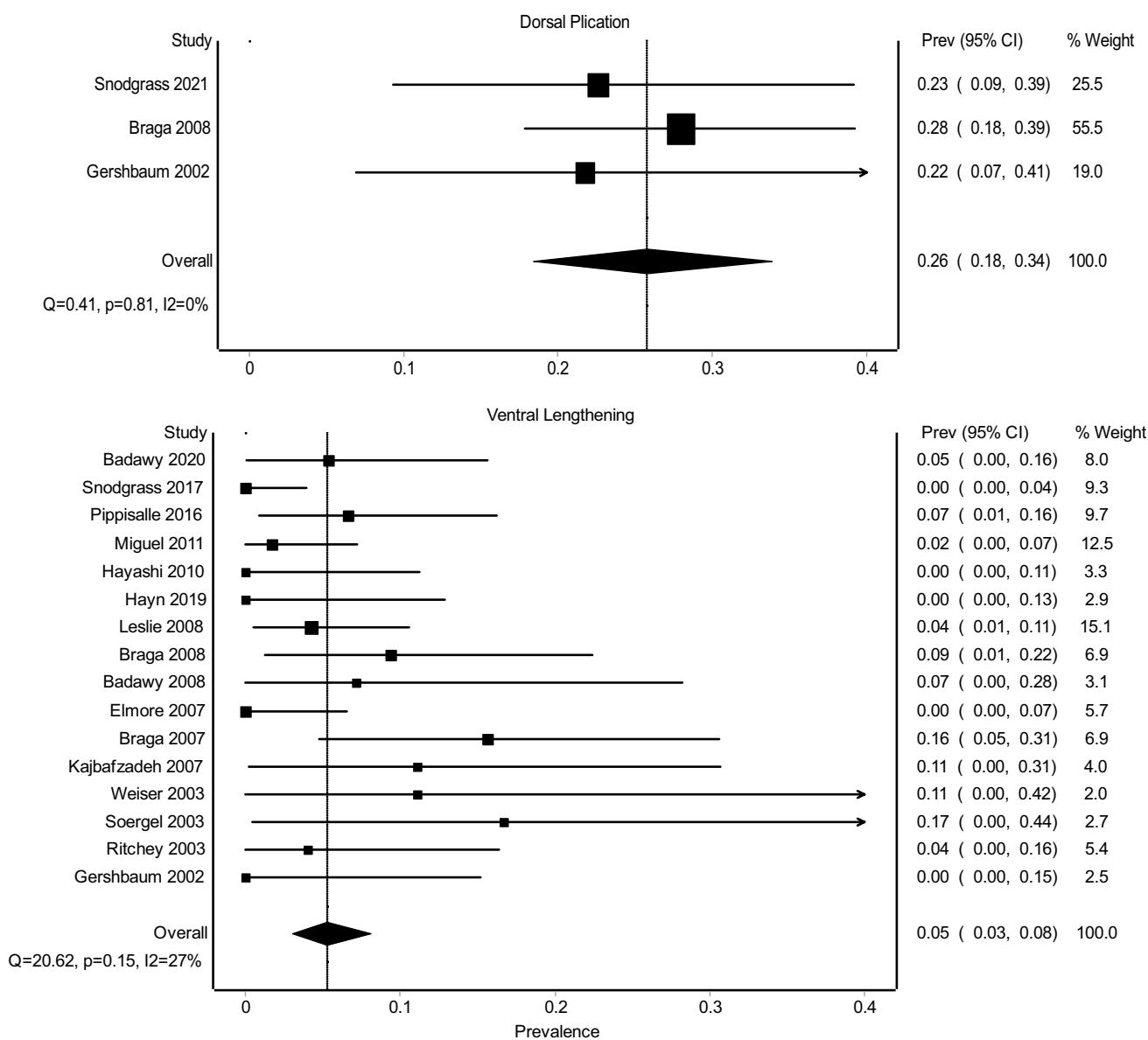


Fig. 3 Forest plot comparing DP and VL techniques. Squares represent % of RC and horizontal line, the 95% confidence interval. The heterogeneity was low in both groups

meta-analysis reported a follow-up of 5–10 years, adolescent outcomes were hardly reported. Husmann [41] raised concerns regarding possibility of erectile dysfunction in patients undergoing urethral plate division or VL procedures. In the present meta-analysis, we have not looked at the incidence of erectile dysfunction following these penile straightening procedures, because it was not reported by many authors. Future research should focus on whether this can be reduced by choosing one VL procedure over the other.

One of the important limitations of this study is variation in urethroplasty techniques. Some authors performed urethral plate elevation [17] and ventral incisions along with a single stage repair and we had to exclude those numbers.

Some added DP to this step to prevent ventral shortening again [13–16] and we had to exclude them as well. Some papers on VL reported both single stage urethroplasty and staged procedure [21, 23, 29] following urethral plate division. Thus the surgeon’s preference over hypospadias correction technique was highly subjective and prone to bias. Despite these limitations, as we excluded papers with high ROB we had pure data on either DP alone or VL alone to compare RC rates.

Documentation of finer details at primary procedure and strict follow-up up to adulthood is the only way to gain more knowledge on this subject over the years. While some authors have mentioned that penile length is shorter

Table 1 Outcomes during follow-up after different types of chordee correction

Author	Type of repair	Chordee correction method	Number	Recurrent chordee (%)	Mean follow-up (months)
Snodgrass 2021 [17]	Proximal TIP	Dorsal Plication (DP) (midline)	31	7 (22.6%)	44
Badawy 2020 [18]	STAG repair	Urethral plate transection + Ventral corporotomies	37	2 (5.4%)	38
Snodgrass 2017 [19]	STAG repair	Urethral plate transection + Ventral corporotomies	43	0	22
Pippisalle 2016 [20]	Staged foreskin flap/graft	Ventral corporotomy (DTITA)	45	3 (6.7%)	29
Miguel 2011 [21]	Staged foreskin flap/single stage foreskin tube	SIS corporoplasty	58	1 (1.7.5)	50
Hayn 2019 [22]	Staged foreskin flap	SIS corporoplasty	13	0	36
Hayashi 2010 [23]	Single stage foreskin tube or Staged foreskin flap	TV flap corporoplasty	15	0	12
Leslie 2008 [24]	Staged foreskin flap	Dermal graft corporoplasty	29	1 (3.4%)	24
		TVFG corporoplasty	21	0	
		SIS corporoplasty	20	2 (10%)	
Braga 2008 [25]	Transverse prepucial flap urethroplasty	Dorsal plication	68	19 (27.9%)	62
		TVF corporoplasty	32	3 (9.3%)	
Badawy 2008 [26]	Staged foreskin flap	Dermal graft corporoplasty	14	1(7%)	120
Elmore 2007 [27]	Staged foreskin flap	SIS corporoplasty	26	0	38
Braga 2007 [28]	Transverse prepucial flap urethroplasty	Dura graft corporoplasty	9	4 (44%)	63
		TVF corporoplasty	23	1 (4.3%)	
Kajbafzadeh 2007 [29]	TIP/Tubed prepucial island flap	TVFG corporoplasty	18	2 (11%)	16
Weiser 2003 [30]	Staged foreskin flap	SIS corporoplasty	9	1 (11%)	16
Soergel 2003 [31]	Staged foreskin flap	SIS corporoplasty	12	2 (16%)	12
Ritchey 2003 [32]	Staged foreskin flap	SIS corporoplasty	3	0	24
		Dermal graft corporoplasty	3	0	
		TVFG corporoplasty	19	1(5.3%)	
Gershbaum 2002 [33]	Thiersh Duplay Single stage	Dorsal plication	23	5 (21.7%)	120
	Staged foreskin flap	TVFG corporoplasty	11	0	

Table 2 Pooled outcomes of recurrent chordee (RC) during follow-up after different types of chordee correction

	Total	RC (%)	95% CI	P value
Dorsal plication versus ventral lengthening				
Dorsal plication	122	31 (25.4%)	18%–33%	0.00001
Ventral lengthening	460	24 (5.3%)	4%–8%	
Dorsal plication versus ventral lengthening subtypes				
Ventral corporotomy	125	5 (4%)	1%–9%	0.00001
Ventral TVF/TVFG corporoplasty	139	7 (5%)	2%–10%	0.00001
SIS corporoplasty	141	6 (4.2%)	2%–9%	0.00001
Dermal/dura graft corporoplasty	55	6 (10.9%)	5%–22%	0.028
Among ventral lengthening subtypes: TVF vs. TVFG corporoplasties				
Tunica vaginalis flap (TVF)	70	4 (5.7%)	2%–14%	1
Tunica vaginalis free graft (TVFG)	69	3 (4.3%)	1%–12%	
Among ventral lengthening subtypes: 1 layer vs. 4-layer SIS corporoplasties				
1 layer SIS corporoplasty	90	1 (1.1%)	0 – 6%	0.02
4-layer SIS corporoplasty	51	5 (9.8%)	4%–21%	

Table 3 Studies on recurrent chordee management describing type of primary chordee repair, possible cause of recurrence & secondary management

Author; year	Total number of recurrent chordee	Initial chordee correction technique			Cause of recurrent chordee	Recurrent chordee management	
		Dorsal plication	Ventral lengthening	Others		One stage	Staged repair
Abosena 2020 [34]	59	43	3 (dermal graft)	13 (unknown)	Skin contracture and peri urethral fibrosis 32	Dorsal plication/skin detethering 32 Urethral mobilization corporal graft 12	Excision of scarred urethral plate and dermal graft 15
Snodgrass 2019 [35]	56	23	15 (13 graft)	18 (chordee excision)	Skin scar 4 Short urethra 8 Corporal disproportion 42	–	Ventral corporotomies (Straightening and closure) 56
Hanna 2017 [36]	45	30	15 (dermal graft)	–	Skin/subcutaneous scarring 27	Dorsal plication 12 + Buccal mucosa graft (BMG) inlay 23	BMG graft 11
Yucel 2016 [37]	10	4	–	6 (unknown)	Skin/ventral scar 10	Skin detethering + Dorsal plication 10	–
Total	170	100 (59%)	33 (19%)	37 (22%)	Skin/subcutaneous scar 73 (43%)	Skin detethering + Dorsal plication 54 (31%)	

in hypospadiac children [42] others have reported it to be normal [43]. Special attention to assess penile length, both dorsally and ventrally before and after surgery, could help assess this important patient centric aspect. Hayashi [23] felt 15% VC was insignificant, while in a survey by Bologna [40] up to 20% chordee was considered insignificant. Bologna [40] reported that VL was opted only for chordee above 50% while Schlomer [44] recommended VL for chordee above 45%. This approach has changed of late and now more surgeons are opting for VL for chordee above 30–40% [19, 26]. Hypospadiologists should introspect whether they are under-estimating the degree of VC and overdoing DP in an overzealous attempt at a single stage procedure. In this regard the authors support artificial erection test and objective measurement of VC with a goniometer before taking the major decision on the type of VC correction technique during primary hypospadias repair.

Conclusions

For correction of ventral chordee during primary proximal hypospadias repair, dorsal plication carries a higher risk of recurrence compared to ventral lengthening procedures. It is essential to objectively measure the extent of chordee and

choose the ideal VC correction technique based on the best available evidence.

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Declarations

Conflict of interest Nil.

Ethical approval IRB exempted.

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