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ERECTILE DYSFUNCTION (DK MONTAGUE, SECTION EDITOR)

Congenital Penile Curvature: Update and Management

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Abstract Congenital penile curvature results from disproportionate development of the tunica albuginea of the corporal bodies and is not associated with urethral malformation. Patients usually present after reaching puberty as the curvature becomes more apparent with erections, and severe curvature can make intercourse difficult or impossible, at which point surgical repair is recommended. Excellent outcomes can be expected with surgical intervention. The three most commonly used repair techniques are the original Nesbit procedure, modified Nesbit procedure, and plication. Nesbit and modified Nesbit techniques require that an incision is made in the tunica albuginea while plication techniques utilize plicating sutures without an incision. While Nesbit and modified Nesbit techniques are more complex operations, these generally result in less recurrences and more satisfactory outcomes as opposed to the quicker and simpler plication technique.

Keywords Congenital chordee · Congenital penile curvature · Nesbit · Congenital penile anomalies · Penile plication · Erectile dysfunction

Introduction

Congenital penile curvature (CPC) or chordee is a relatively uncommon condition that may present in late adolescent or early adult life [1–3]. The incidence is estimated to be 0.6 % [4], but the incidence of clinically significant CPC is much lower, as the degree of curvature and sexual dysfunction varies widely [5]. CPC can be mistakenly classified as Peyronie's disease because the physical manifestations may be similar, yet the etiology and pathophysiology are different. CPC may be ventral, dorsal, or lateral. While dorsal curvature is often associated with epispadias, ventral curvature is often associated with hypospadias. Typically, isolated cases of CPC occur ventrally with an orthotopic meatus. This review will discuss isolated CPC without associated urethral plate malformation such as hypospadias, epispadias, or shortened urethral plate.

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Historical Descriptions

Penile curvature was first noted as early as 130–190 AD by Galen, a physician to Roman gladiators [6]. In addition, it was documented in 1547 that King Henry II of France was not able to father children until his penile curvature was corrected by surgeon Jean Fernel [7,8]. The first documented medical description of congenital penile curvature (CPC) was in 1842 [9], but this condition did not become widely accepted in the medical community until 1967 [10].

Embryology and Classification

As mentioned above, it is important to understand the pathophysiology of CPC as it differs from Peyronie's disease and other congenital conditions causing curvature. Phallic development begins with the appearance of the genital tubercle at 4 weeks gestation [11]. Complete development of the distal urethra, frenulum and circumferential prepuce is dependent upon ventral migration of the urethral and preputial folds [12]. The distally located lacunar folds fuse to form the distal third of the glanular urethra, which is lined

only by ectoderm [12]. All of these infoldings occur in a carefully orchestrated sequence in a proximo-to-distal fashion [13]. Development of the corpora cavernosae via coalescence of the mesoderm that flanks the urethral groove dorsolaterally and is thought to occur secondary to urethral development, possibly acting as an inductive force on development of the erectile bodies [14]. While the exact mechanism that drives penile (corporal and urethral) elongation has not been elucidated, paracrine testosterone production during the second and third trimesters plays a very important role in this process [15]. Deviation in the above signaling pathways can result in CPC.

Devine and Horton [16] classified CPC without hypospadias based on the embryological malformation causing the curvature. Type I is the most severe defect and has thin urethra with deficiency of corpus spongiosus at the site of maximal curvature distally. In addition, there is thick scar tissue ventral to the spongiosum causing the curvature. Type II and III have normal corpus spongiosum, but type II has abnormal Dartos and Buck's fascia and type III has only abnormal Dartos fascia and may be associated with penile torsion. It is important to point out that Type I, II, and III defects are due to abnormal development of the ventral penile structures; therefore, they will have an orthotopic urethral meatus with ventral penile curvature. Type IV was later described by Kramer et al. [17], and is due to corporal disproportion. Also, there have been a few reports of a shortened urethra causing CPC and this was classified as type V curvature [18,19].

Tunica albuginea is mainly composed of collagen bundles and elastic fibers and their symmetric distribution is responsible for the symmetry of erection. Currently, theory holds that the curvature is due to increased elasticity of tunica albuginea on the greater curvature, as demonstrated by the disorganized pattern of collagen fibers on that side [20]. Interestingly type IV congenital curvature has been noted to be associated with penile length of two or more standard deviations above the normal length for that age [21]. Thus, true CPC appears to be a problem of increased elasticity. While the reason for the curvature has been indentified, the embryologic mechanism of improper development of the greater curvature is still unknown. Catuogno and Romano [22] studied the effects of androstanolone on treatment of CPC and found that there is a discrepancy in the androgen receptor expression on the greater and lesser curvature.

Presentation and Preoperative Considerations

While most penile malformations present in childhood, isolated CPC tends to present in late adolescence or early adulthood [1–3]. Patients may be concerned with the aesthetic or functional aspects of their penis. As with any urologic condition, the evaluation begins with a thorough history and physical examination. The patient should be questioned on any previous urologic history, including circumcision. Patients should be asked about the quality of their erections and the directions and degree of curvature. Kelami [23] reported in their series of patients with CPC that ventral deviation occurred in 48 %, lateral deviation in 24 %, dorsal deviation in 5 %, and a combination of ventral and lateral deviation in 23 % of patients. These proportions appear to be similar across other cohorts studied to date [1,2,24•]. The angle of the curvature can range from minimal to very severe deviation. Various angles of curvature have been reported and most patients treated range from 30° to 90° [2,3]. In Kelami's [23] cohort, 16 % of patients had over 60 % deviation, 44 % had deviation between 30 % and 60 %, and 40 % of the patients had less than 30 % deviation [23]. We and others have observed that patients with curvature of 30° or more will seek repair [25]. Proper documentation of deviation direction and angle is important for operative planning. Many authors suggest that photo documentation of the erect penis be included in the preoperative evaluation.

Preoperative counseling should include a full discussion of the operative procedure, potential complications, and especially patient expectations. All of the current straightening techniques result in overall shortening of erections. Studies suggest that an average loss of about 2.5 cm can be anticipated depending on degree of curvature and type of repair used [26]. It is imperative to counsel patients on the loss of length and that most patients are satisfied with the repair despite the decrease in length. Additionally, postoperative expectations and potential complications include palpable sutures that may or may not be bothersome to the patient, postoperative hematoma formation, and glans paresthesias [1,27••]. Changes in penile temperature sensation and vibratory sensation also have been reported [28]. Residual curvature requiring reoperation or recurrence rate differ based on method of repair used and range from 0 % to 48 % [27••, 29]. In the immediate postoperative period, erection suppression has been advocated to prevent suture rupture and/or hematoma formation [30]. Erectile dysfunction (ED) is very rarely reported as a postoperative complication, with rates of de novo postoperative ED in the 1 %–8 % range depending on patient population and technique utilized [29,31]. We typically demonstrate the stretched penile length to the patient as this has been shown in other studies to estimate the erectile penile length postoperatively (see below).

A few studies have examined the psychological impact of repair. Cavallini and Coracciolo [26] reported no significant change in interpersonal relationships before and after the repair, but nearly one quarter of those patients suffered from psychogenic ED that did not improve after surgery. Tal et al. [32••] reported improvement in sexual relationships, overall relationship, and confidence scores after the repair as measured

by Self Esteem and Relationship (SEAR) questionnaire; however, International Index of Erectile Function (IIEF) scores were unchanged. These contradictory findings highlight a need for standardized, validated, prospective outcomes studies for this patient population.

Outcomes after repair vary depending on series, but most report 71 %–100 % satisfactory results [1,27••]. As expected, the definition of "satisfactory result" varies across studies, ranging from anatomic and cosmetic results only to more qualitative outcomes. Due to the risk of complications and understanding that lesser degrees of curvature are unlikely to be clinically significant for function, most authors reserve surgery for patients with curvatures of greater than 30° and those that complain of functional concerns associated with curvature.

Techniques

The repair of penile curvature has evolved over time. Initially, complex urethral mobilization surgery was advocated but is now accepted as being unnecessary in this population. Nesbit [33] was the first to describe plication of the tunica albuginea in 1965. Since then, a number of modifications have been made in attempt to improve outcomes and minimize surgical risks. Additionally, grafting techniques have been studied in this setting where penile length preservation is a concern, but are not currently considered to be standard of care in repair of CPC [34].

Nesbit Technique

Most surgical corrective procedures start with degloving (for circumcised men) and artificial erection. Complete penile degloving is not always necessary and some authors reported satisfactory results using small incisions at the site of maximum curvature (Fig. 1a, b) [3]. The Nesbit technique was initially described as an excision of horizontal ellipses of the tunica albuginea on the greater curvature [33]. Depending on the location of the point of maximal curvature along the greater curvature, the neurovascular bundle may require mobilization off the tunica albuginea. This can be accomplished via a lateromedial approach or via excision of a segment of superficial dorsal vein and medial-lateral mobilization as described by Giammusso et al. [33,35]. Allis clamps are then applied until the desired straightening is achieved. If ventral-dorsal and lateral curvature need to be corrected, multiple plications may be needed to straighten the erection in all dimensions [33,36]. These ellipses can be excised and closed transversely in a stepwise fashion until curvature is completely corrected.

Satisfactory results after Nesbit plication (reported as either overall patient satisfaction or success of repair not requiring any further intervention) ranged from 80 % to 100 % (Table 1) [1,23,26,27••, 29,36,37]. Reoperation or recurrence of curvature was observed in 0 % to 8 % of the patients [3,23,26,27••, 29,36,37], with one study reporting 33 % recurrence of curvature [1]. Mean operative times for Nesbit procedure reported by Rolle at al. [37] were 62±15 min [37]. Other reported complications rates include glans paresthesias (2 %–75 %) [1,26,27••, 36,37], penile shortening (0 %–50 %) [1,21,26,27••, 37], and palpable sutures or suture granulomas (0 %–100 %) [1,27••, 29,36]. Occasional wound infection and hematoma formation also have been reported as early postoperative complications [26,29,36]. Postoperative ED was reported in about 1 % of patients [29].

Modified Nesbit Technique

In 1973, Saalfeld et al. [38] proposed a modification to Nesbit procedure that consisted of a longitudinal incision in the tunica with transverse closure. This method is based on the idea that making a longitudinal incision as opposed to transverse incision over the corpora will minimize risk of injury to the neurovascular bundle as well as shorten the procedure by minimizing the need for more extensive lateral dissection (Fig. 1c, d) [39]. Yachia [39] further modified this approach with simple longitudinal incisions made along the greater curve and closed in a horizontal fashion (Fig. 1c, d). The length of the incision is limited to about 1 cm to prevent excessive indentation of the penis [40].

Success rates of this procedure have been reported to be 80 % to 100 % (Table 2) [1,3,35,40–43]. As with all straightening operations, penile shortening is the most commonly reported postoperative complaint, with rates ranging from 9 %–67 % versus 5 %–50 % with Nesbit procedure [1,3,35,40,42]. None of the patients reported permanent paresthesias, but did have transient changes in sensation (0 %–14 %), thus suggesting that less mobilization of the neurovascular bundle utilized in this modification may indeed result in less sensory loss [1,3,42,43]. As stated previously, correcting ventral deviation requires intervention on the dorsal aspect of the penis and special care has to be used in dissecting the dorsal neurovascular bundle. Utilizing this approach, satisfaction rates usually exceeded 90 % with very few complications reported.

Plication

In 1985, Essed and Schroeder [44] proposed that incisionless placation of the tunica albuginea, thus minimizing the risk of injury to neurovascular bundles as well as decreasing the complexity and time of the procedure. Interestingly, Nesbit [33] did initially perform this procedure in 1965,

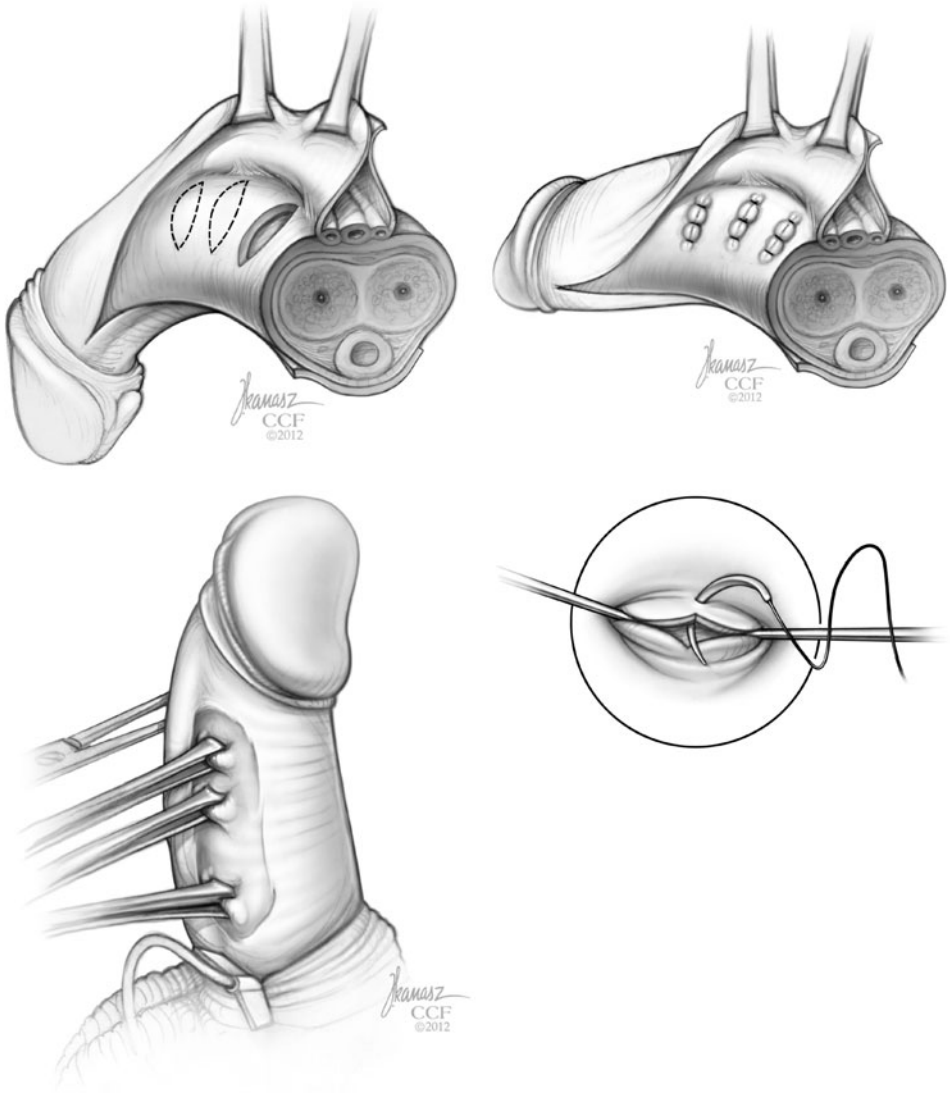

Fig. 1 Classic Nesbit procedure **a**, **b** demonstrating mobilization of Buck's fascia and horizontal elliptical excisions along the greater curvature. **c** In the Yachia modification, Allis clamps are applied to straighten the erection, and a **d** longitudinal incision is made and closed horizontally in each clamp location

but found that the patient had recurrence of curvature within 6 months of surgery, and thus abandoned this technique. After degloving and artificial erection, Allis clamps are used on the greater curvature to estimate the number and length of tissue needed for plications, or plicating sutures are placed and adjusted as needed to achieve optimal straightening. The artificial erection is then released and sutures tied [27••, 44,45].

Some studies have reviewed the outcomes of plication methods for repair of CPC [1,27••, 31,45–50]. Most of these are small cohort studies, and some included patients with Peyronie's disease. In this review, we have made our best

Table 1 Nesbit technique: summary of studies

Study	Patients, *n*	Age, *y*	Angle, *degree*	Successful, *%*	Shortening, *%*	Sensation changes, *%*	Suture, *%*	Hematoma, *%*	Recurrence/ reoperation, *%*
Poulsen and Kirkeby [29]**	95	19*	NA	86	NA	NA	0	4	8
Andrews et al. [36]	106	25	15–90	96	5	2	3	1	5
Rolle et al. [37]	32	22	40–90	100	0	40	NA	NA	0
Cavallini and Caracciolo [26]**	56	24	> 25	80	20	2	NA	NA	NA
Nyirady et al. [1]	18	24	30–90	89	17	6	17	NA	33
Leonardo et al. [27••]	12	17	> 30	100	50*(slight)	75	100	NA	0

NA not available

* Median ** Technique was assumed to be the original Nesbit procedure; was not clearly stated in methods to determine otherwise

Table 2 Modified Nesbit technique: summary of studies

Study	Patients, *n*	Age, *y*	Angle, *degree*	Successful, *%*	Shortening, *%*	Sensation changes, *%*	Suture, *%*	Hematoma, *%*	Recurrence/ reoperation, *%*
Sassine et al. [43]*	32	26	30–90	95	NA	0	NA	0	2
Giammusso et al. [35]*	12	30	NA	100	67	7 (maintaining erection)	25	NA	0
Ghanem and Shamloul [42]	45	27	> 30	98	9	2	0	NA	7
Daitch et al. [40]	5	22	40–90	80	60	NA	NA	NA	NA
Nyirady et al. [1]	62	24	30–90	96	29	0	86	NA	43
Popken et al. [3]	105	23	30–100	95	25	14	30	11	7

NA not available; *CPC* congenital penile curvature

* Mixed cohort: includes CPC and Peyronie's disease

attempt to include outcomes data for patients with CPC only. Satisfactory outcomes were reported in 71 %–100 % of the patients whether it was based on patient satisfaction or successful straightening (Table 3) [1,2,24•, 27••, 29,31,45–48,50] with one study showing only 35 % success rate [29]. Recurrence or reoperation rates after repair ranged from 1 % to 48 % [1,2,27••, 45–48]. Palpable sutures (9 %–73 %), sensory changes that sometimes were transient (0 %–37 %), and minimal penile shortening that did not appear to affect satisfaction (16 %–74 %) were reported as the most common complaints associated with plication repair [1,2,24•, 27••, 29,31,45–48,50]. The main concern about this repair continues to be durability.

A few modifications to plication technique have been proposed to address and minimize some of the complications. One of these is the Lue 16-dot plication method or the multiple parallel plications technique [51]. This technique employs multiple nonabsorbable sutures placed at the point of maximum curvature of the convex side. These are placed opposite to each other either between the deep dorsal vein and dorsal arteries for ventral curvature repair or on either side of the corpus spongiosum for dorsal curvature repair [52,53]. The main advantages of this technique are short procedure time, ability to perform procedure under local anesthesia, and no need for dissection of the neurovascular bundle. Additionally, since albuginea is the thickest at the 5, 7, and 12 o'clock positions [54], these are optimal positions for suture placement to prevent tearing through the tunica. Disadvantages include palpable sutures and penile shortening. Success rates for this method are 80 %–93 % [51,53]. Major complications included complaints of shortening of the penis (5–15 mm, 41 %), bother from suture knots (12 %), pain with erection (11 %), narrowing or indentation of the shaft of the penis (9 %), decreased sensation (6 %), and hematoma formation (4 %) [51]. The mean reported operative time was 45 min and 85 % of the cases were done under local anesthesia only [51].

Nonabsorbable sutures have been classically used for penile curvature plication repair techniques to provide tension necessary for straightening the penis and preventing recurrence. One of the main complaints in the penile curvature repair surgery is patient's ability to palpate the sutures. Therefore Hsieh et al. [30] suggested use of absorbable sutures (2-0 Vicryl [Ethicon, Inc., Somerville, NJ]) in 2001. He reported no complaints, palpable suture knots, or suture granulomas after repair and 82 % were satisfied; 87 % had less than 15° of residual curvature [2]. However, 28 % of the patients experienced suture failure. Basiri et al. [24•] compared outcomes of plication technique using 2-0 Vicryl and nylon suture in a cohort of 35 patients and showed similar correction rates in two groups (88.2 % vs 88.9 %) but significantly lower rate of palpable sutures (39 % vs 6 %) when using Vicryl as opposed to nylon sutures respectively [24•].

Comparison of Techniques

Recently, a number of retrospective reviews compared the outcomes of Nesbit/modified Nesbit procedure to the plication procedures and showed that while there is decreased risk of complications and loss of sensation, there is an increased risk of recurrence associated with plication techniques [1,27••, 29]. Two of the three studies also reported overall better outcomes in patients treated with Nesbit or modified Nesbit technique.

In 1995, Poulsen et al. [29] reported outcomes in 118 patients (95 treated with Nesbit procedure and 23 treated with plication) who presented for follow up after 3–5 months. There was a drastic difference in overall satisfactory outcomes with 86 % good or acceptable results in Nesbit group as compared to 35 % in the plication group. While only 8 % of patients in the Nesbit group required reoperation, 48 % of those in the plication group did. They found more early complications in the Nesbit group such as a 4 % rate of hematoma formation; however, none were observed in the plication group. They also reported one case of decreased erectile function in the Nesbit group and scar formation as some of the late complications; in the plication group, late complications were associated with palpable sutures.

Table 3 Plication technique: summary of studies

Study	Patients, n	Age, y	Angle, degree	Successful, %	Shortening, %	Sensation changes, %	Suture, %	Hematoma, %	Recurrence/ reoperation, %
Nooter et al. [48]	22	26	20–90	95	36	0	NA	NA	5
Poulsen and Kirkeby [29]	23	21*	NA	35	NA	NA	9	0	48
Hauck et al. [46]	23	23	>30	82	65	0	9	0	26
Chien and Aboseif [45]	22	39	>90	95	18	NA	NA	9	NA
Van Der Horst et al. [50]	22	NA	30–90	77	NA	9	5	NA	5
Lee et al. [47]	68	21	30–90	97	38	4	51	NA	1
Paez et al. [31]	26	31	NA	73	NA	27	54	NA	NA
Nyirady et al. [1]	7	24	30–90	71	16	2	6	NA	2
Leonardo et al. [27••]	19	22	> 30	100	74	37	74	NA	16
Basiri et al. [24•]	35	34	55	89	20	NA	23	NA	NA
Hsieh et al. [2]	114	24	30–90	82	25	16	20	NA	14

NA not available

* Mixed cohort: includes CPC and Peyronie's disease

Nyirady et al. [1] compared outcome in 87 patients treated with three different techniques: Nesbit (n=18), plication (n=7), and modified Nesbit (n=62). Patients were followed for a mean of 89 months and overall 97 % were successfully treated. Shortening of the penis was reported by 16 % of patient with Nesbit or modified Nesbit procedure versus 28 % of the patients after plication. On the other hand, no patients in the plication group complained of altered penile sensation as opposed to 5 % in the Nesbit group and 1 % in the modified Nesbit group. Patients in the plication group had an increased number of recurrences and were more likely to complain of palpable sutures. Overall satisfaction was reported to be 88 % in the Nesbit group, 98 % in the modified Nesbit group, and 71 % in the plication group.

In 2012, Leonardo et al. [27••] studied outcomes of 31 patients with CPC with a mean follow up of 39 months. Overall, they achieved 100 % satisfaction with both techniques. Penile shortening was reported by 50 % of the patients in the Nesbit group as opposed to 75 % in the plication group, and despite the fact that sutures were palpable in both groups, one of the main complaints associated with plication as compared to Nesbit operation was patients' discomfort and ability to feel the sutures during erection (21 % vs 0 % respectively). On the other hand, 75 % of patients undergoing Nesbit procedure reported paresthesias of the glans as compared to 37 % in plication group [27••].

Length-Preserving Techniques

It is also important to note that patients with isolated penile curvature tend to have above average penile length [21] as compared to those with more complex curvatures associated with hypospadias, and thus, loss of length is often not a functionally significant complication. Factors that affect loss of length in CPC repair techniques have been shown to be the degree of the curvature and direction of the curvature, specifically ventral or ventrolateral curvature [55]. Thus, patients with severe curvature or those where shortening of the penis would be unacceptable may benefit from alternative means of repair. This should be done only in special circumstances, however, because the length of operation, recovery, and complications such as de novo ED may be increased with more complex repairs. Grafts used specifically in CPC have included dorsal vein [56,57], tunica albuginea [58], tunica vaginalis [59], and dermis [60,61] among others (also off-the-shelf products such as subintestinal submucosa (SIS) and bovine pericardium, among others). When tunica albuginea is used for grafting, it is excised from the greater curvature similar to the Nesbit procedure and implanted in an incision on the lesser curvature [58]. Authors reported a 50 % reduction in shortening of the penis associated with this technique and a 93 % satisfaction rate in a mixed cohort of acquired and congenital curvature patients. Simonato et al. [60] used dermal grafts and reported no change in penile length in 27 % of the patients and an in increase in length in 73 % [60]. Additionally, authors reported mean operative time of 130 min for dermal grafting [60] as compared to 45 min for plication technique [51]. Some of the specific complications of the grafting techniques include decreased rigidity of erections due to venous leak or graft contraction resulting in recurrent curvature, and glans paresthesias [60,61]. Grafting techniques have been studied more extensively in the setting of Peyronie's disease and are reviewed elsewhere [62].

Conclusions

CPC is a result of abnormal development of the tunica albuginea of the corporal bodies and increased elasticity of

the tunica on the greater curvature. Patients usually present for repair in their 20s while the curvature becomes apparent with progression of puberty. The severity and direction of the curvature can range with most patients seeking intervention for curvatures greater then 30°. A number of techniques have been described that can be used to repair with overall success rates ranging from 71 % to 100 %. The Nesbit repair is a successful and durable repair with success rates ranging from 80 % to 100 % (median 92.5 %) and all but one cohort reporting recurrence rates or reoperation rates of <10 % [1,23,26,27••, 29,36,37]. Since this procedure requires an excision of one or more ellipses from the tunica albuginea, thus increasing risk of damage to neurovascular structures, it has been associated with often transient sensory changes in 2 %–75 % of patients. One of the most popular modifications to the Nesbit procedure was the Yachia modification, which employs longitudinal incision in the tunica with transverse closure as opposed to excision of an ellipses of the tunica to avoid damage to neurovascular structures. This is our preferred method of repair, with success rates ranging 80 %–100 %, median 95.5 % with sensory changes reported in only 0 %–14 % of patients [1,3,35,40,42,43]. Finally, plication was proposed as a simple and quick method of repair that avoids making an incision in the tunica albuginea all together. The success rates are lower for this method (35 %–100 %, median 82 %) and rates of recurrence or reoperation are higher (2 %–48 %) [1,2,24•, 27••, 29,31,45–48].

Overall, patients with CPC can expect excellent outcomes with repair and minimal side effects. Risks and benefits vary depending on the chosen method of repair should be discussed with patient pre-operatively to ensure appropriate patient expectations. Widely varied outcomes highlight the need for standardized outcomes measures reporting and there is a need for long-term follow up and outcome reporting to better delineate differences in techniques.

Disclosures No potential conflicts of interest relevant to this article were reported

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- •• Of major importance

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