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Correlation of severity of penile torsion with type of hypospadias & ventral penile curvature and their management



A. Bhat^{a,*}, K. Sabharwal^a, M. Bhat^b, M. Singla^a, R. Upadhaya^a, V. Kumar^a

^a Department of Urology, S.P. Medical College, Bikaner, Rajasthan, 334003, India

^b Department of Preventive and social Medicine, S.P. Medical College, Bikaner, Rajasthan, 334003, India

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KEYWORDS

Congenital anomalies; Hypospadias; Penile torsion; Chordee; Correlation of penile torsion and chordee; Mobilization of urethral plate and spongiosm; Management of penile torsion

Abstract

Objective: This study was carried out to evaluate the correlation between the severity of penile torsion on the one hand and the degree of ventral penile curvature and the type of hypospadias on the other. We also assessed the effectiveness of correction of chordee and torsion by penile degloving and mobilization of the urethral plate and the corpus spongiosum.

Patients and methods: This prospective study included 116 patients with hypospadias and penile torsion out of a total of 376 primary hypospadias cases seen between January 2006 and June 2013. The patients' age ranged from 8 months to 26 years with a mean age of 8.37 and a median age of 6.4 years. Prior to surgery the type of hypospadias as per location of the meatus, the presence or absence of chordee, the size of the dorsal hood and deviation of the median raphe on the dorsal hood were noted. The torque of the penile shaft (torsion) toward either side of the midline and ventral curvature was measured using a sterile small protractor around the penile shaft. The techniques used for the correction of penile torsion and chordee were penile degloving and mobilization of the corpus spongiosum with the urethral plate and the urethra. *Results:* The abnormal penile rotation ranged from 15° to 110° (average 51.98°). In 70.69% of the patients the torque was on the left side, while it was on the right in 29.31%. 11.2% of the patients had a severe torque, while it was moderate in 37.94% and mild in 50.86% of the cases. The mean torque was $62.38^{\circ} \pm 23.03^{\circ}$ in patients with distal penile (80 cases), $38.04^{\circ} \pm 18.50^{\circ}$ in patients with mid penile (24 cases) and $18.25^{\circ} \pm 3.33^{\circ}$ in patients with proximal penile hypospadias (12 cases) (*P* value = 0.001). Ventral curvature was seen in 71 cases. Mean ventral curvature was $38^{\circ} \pm 18.55^{\circ}$, $44.28^{\circ} \pm 21.11^{\circ}$ and $73.58^{\circ} \pm 32.96^{\circ}$ in patients with distal penile, mid penile and proximal hypospadias, respectively

* Corresponding author.

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E-mail addresses: amilalbhat@rediffmail.com (A. Bhat), drkvsabharwal@gmail.com (K. Sabharwal), mahak199027@gmail.com (M. Bhat), drmanishsingla@gmail.com (M. Singla), surgeon.ups@gmail.com (R. Upadhaya), vinaysinghkgmc99@gmail.com (V. Kumar). Peer review under responsibility of Pan African Urological Surgeons' Association.

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(*P* value = 0.001). The procedures of choice for the repair of penile torsion were penile degloving in 11% of the cases, mobilization of the urethral plate and the corpus spongiosum in 28% of the cases, mobilization of the proximal urethra in 40% of the cases and mobilization of the urethral plate into the glans in 21% of the cases. Chordee could be corrected using penile degloving and mobilization of the urethra/urethral plate in all cases.

Conclusions: Penile torque is more common and severe in distal hypospadias, while ventral curvature is seen more often in proximal hypospadias. The degree of torsion is inversely proportional to the severity of ventral curvature. Techniques for the repair of penile torque and ventral curvature include penile degloving and mobilization of the urethral plate with the corpus spongiosum and the urethra.

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Introduction

Penile torsion is a congenital deformity with helical rotation of the corporal bodies of the penile shaft, while the proximal parts of the corporal bodies remain fixed at the attachment with the pubic rami. The usual direction of penile torsion is counterclockwise (i.e. toward the left) with the urethral meatus in oblique position [1,2]. The median raphe may extend in a spiral manner from the base of the penis ventrally and around the penile shaft. Depending on the degree of rotation from the midline, torsion may be classified as mild ($<45^\circ$), moderate ($45-90^\circ$) and severe ($>90^\circ$) [3]. The incidence of isolated penile torsion has been reported to be 1.7% to 27%, that of severe torsion to be 0.7% [1,2]. Torsion is also found to be associated with epispadias, hypospadias and chordee without hypospadias. Penile torsion with hypospadias is an overlooked entity the true incidence of which is unknown. It is commonly associated with anterior hypospadias [4,5]; only one study describes penile torsion with distal hypospadias [5]. While there is no need for treating an isolated penile torque of less than 60°, which primarily presents a cosmetic problem, penile torsion associated with hypospadias and chordee should be corrected. Once a patient is subjected to hypospadias repair, one should be aware of the possible presence of penile torsion and, if present, it should be corrected at the same time. The importance of recognition of torsion lies in the fact that a simple additional maneuver such as penile degloving and re-attaching is all that may be required for the correction of torsion during hypospadias repair. Techniques for the repair of torsion include penile degloving with skin re-attachment [2,4,5], dorsal dartos wrap rotation [3], pubic periosteal stitch [6], untwisting plication sutures [7] and mobilization of the urethral plate and urethra [4,8,9]. The correlation between the degree of penile torsion on one hand and the type of chordee without hypospadias and severity of ventral penile curvature on the other has been reported in only one study so far [4].

Therefore, the objective of this study was to evaluate the correlation between the severity of penile torsion on one hand and the degree of ventral penile curvature and the type of hypospadias on the other and to assess the effectiveness of correction of the chordee and torque by penile degloving and mobilization of the urethral plate and corpus spongiosum.

Patients and methods

Patient selection

A prospective study of 116 patients with penile torsion with hypospadias operated between January 2006 and June 2013 was

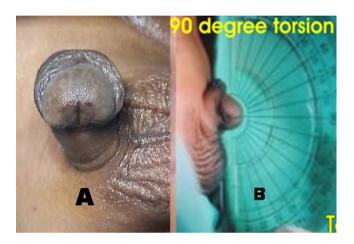


Figure 1 (A) 90° right-sided torsion. (B) Measuring penile torsion with a modified sterile protractor

carried out. Out of a total of 391 cases documented in our hypospadias registry, there were 376 cases with primary hypospadias. The patients' age ranged from 8 months to 26 years with a mean age of 8.37 and a median age of 6.4 years. The patients were evaluated by clinical examination assessing the type of hypospadias as per location of the meatus, the presence or absence of chordee, the size of the dorsal hood and the deviation of the median raphe on the dorsal hood. All the patients were operated by a single surgeon (ALB) with the same type of instruments and similar suture material. Only patients with primary hypospadias who had a penile torque $>15^{\circ}$ to either side were included in the study. Patients requiring redo hypospadias repair and those with perineal or perineo-scrotal hypospadias were excluded. The degree of penile torsion to either side was measured using a small sterile protractor around the penile shaft (Fig. 1). The extent of the median raphe both proximally to the meatus and distally at the dorsal hood was noted (Fig. 2A and B), as well as the degree of deviation of the median raphe from the midline. Institutional Ethical Committee clearance and consent of the parents/patients was taken for data evaluation, photographs and inclusion in study.

Surgical technique

The penile torsion is evaluated by using a sterile modified protractor (Fig. 1) which measures the rotation of the urethral meatus from the midline. The distal extent of the median raphe is marked on the dorsal hood or foreskin. A Gittes test (intracorporeal saline injection and simulated erection) is used to assess the degree of chordee



Figure 2 (A) Deviation of the median raphe starting at the penoscrotal junction. (B) Deviation of the median raphe starting proximally to the peno-scrotal junction

and torque. A circumferential circum-coronal incision is done and extended ventrally in U-shape on the penile shaft along the margins of the urethra, encircling the native meatus and preserving the urethral plate. After creating a dissection plane at the level of Buck's fascia, penile degloving is done down to the root of the penis. The correction of the torsion is evaluated using the Gittes test (Fig. 3D). A dissection plane is created between Buck's fascia and the tunica just proximal to the meatus, then the urethral plate with the corpus spongiosum is mobilized up to the corona, and the correction of the torque is checked again using the Gittes test (Fig. 4E). Next, the corpus spongiosum with the urethral plate is mobilized into the glans up to the future meatus (Fig. 3E), and the correction is measured again using the Gittes test. If torsion persists, the proximal urethra is mobilized up to the perineum, and correction of the torsion is evaluated. In case of still persisting torque, it is adjusted with a skin flap closure. These surgical steps for the correction of chordee and torque have been described in earlier reports [4,8,9]. Tubularized incised plate urethroplasty, spongioplasty and glanuloplasty are done with a urethral catheter left in situ (Figs. 3 and 4). If minimal torque persists, it is corrected by skin flap adjustment. Cephalosporins, analgesics and anti-inflammatory drugs are administered for 7 to10 days as long as the catheter is left in situ.

At the follow-up visits (1, 3, 6 and 12 months postoperatively and then yearly), the patients are subjected to local examination in order to evaluate the outcome of the correction of torsion, and the parents of minors as well as the adult patients are asked about the presence of torque or chordee during morning erections.

Statistical method

The correlation between the mean degree of torsion/chordee \pm SD and the type of hypospadias was analyzed statistically using Student's *t*-test with *P* < 0.05 considered to be significant.

Results

Out of the 376 patients with primary hypospadias operated during the study period, 116 cases had a penile torsion of more than 15° . Distal, mid penile and proximal hypospadias was encountered in 80, 24 and 12 cases, respectively. The torque varied from 15° to 110° with an average of 51.98° . Left-sided torsion (Figs. 3 and 4) was seen in 70.69% and right-sided torsion (Fig. 5) in 29.31% of the patients. Ventral curvature varying from 15° to 125° (average

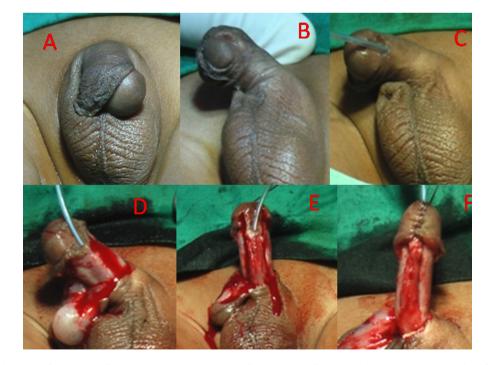


Figure 3 Operative steps of correction of moderate penile torsion. (A)–(C) About 70° left-sided torsion. D. Torsion persisting after penile degloving. (E) Torsion corrected after mobilization of the urethral plate with the spongiosum into the glans and urethra. (F) Incised plate urethroplasty and spongioplasty.

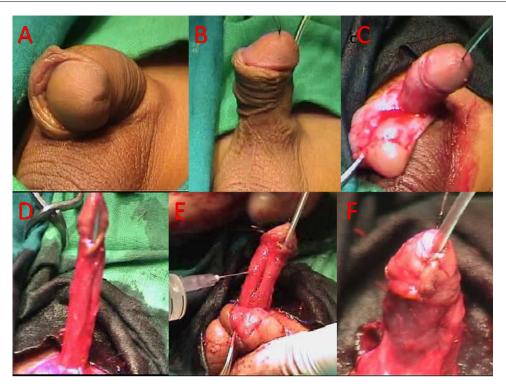


Figure 4 Showing operative steps of correction of severe penile torsion. (A) and (B) More than 90° C. Showing penile torsion persisting after penile de-gloving. (D) Correction of penile torsion after mobilization of urethra and spongiosum. (E) Gittes test to confirm correction of torsion.



Figure 5 Showing more than 90° torsion on right side.

46.58°) was present in 71 cases. The rotation of left-sided torque ranged from 15° to 110° (average 54.9°), while that of right-sided torque ranged from 15° to 95° (average 43.8°) (Table 2a&b). Severe torque (>90°) was seen in 11.2%, moderate torque (45°–90°) in 37.94% and mild torque (15°–45°) in 50.86% of the cases. The torsion was significantly more severe when associated with distal hypospadias as compared to mid penile and proximal hypospadias, i.e. $62.38^{\circ} \pm 23.03^{\circ}$ vs. $38.04^{\circ} \pm 18.50^{\circ}$ and $18.25^{\circ} \pm 3.33^{\circ}$ (P = 0.001) and it was similar when seen separately on left and right side (Table 2c&d). The mean chordee in distal penile hypospadias was $38^{\circ} \pm 18.55^{\circ}$ as compared to $73.58^{\circ} \pm 32.96^{\circ}$ in proximal penile hypospadias (P value = 0.001).

The median raphe was initially located in the midline and then deviated to the opposite side of the torsion in 30% of the cases. In another 30% of the cases the deviation was located at or proximal to the penoscrotal junction, while in 40% the deviation was between the penoscrotal junction and the meatus. The attachment of the median raphe at the dorsal hood was found to be on the contra-lateral side of the torsion in 100, on the same side in 10 and on both sides in 6 cases. The end attachment of the median raphe was at the distal most part of the dorsal hood which was almost equal to the degree of torsion in almost all cases. The deviation of the median raphe, which was toward the right side in 70.69% of the patients, varied from 15° to 110° with an average of 51.98°.

The methods of choice for the correction of the torque were penile degloving in 11%, mobilization of the urethral plate and the corpus spongiosum in 28%, mobilization of the proximal urethra in 40% and mobilization of the urethral plate into the glans in 21% of the cases. Correction of the chordee was possible by penile degloving and mobilization of the urethra/urethral plate in all cases. None of the patients required dorsal plication or division of the urethral plate. Tubularized incised plate urethroplasty, spongioplasty and glanuloplasty were performed in all cases.

The patients were followed up for 12–48 (mean 26) months, and the overall incidence of complications was 7.75%. Residual/recurrent torsion or chordee was not seen in any of the patients. Complications seen in 5 patients (4.3%) with distal hypospadias included infection and disruption in 1 (0.86%), meatal stenosis in 3 (2.6%) and ure-thral stricture in 1 (0.86%). The infection healed spontaneously, while the meatal stenosis and urethral strictures required urethral calibration. One patient (0.86%) with mid penile hypospadias and two patients (1.72%) with proximal hypospadias developed urethral fistulae. Superficial skin flap necrosis which healed with time was seen in one patient (0.86%) with proximal hypospadias. Secondary surgical intervention in the three patients with urethral fistulae and the one with urethral disruption was successful.

Discussion

Penile torsion in association with hypospadias is underreported with only 109 cases reported in literature [4,5,10,11]. In this prospective study, we report on 116 cases with a torsion >15°. A torsion of less than 60° does not usually require treatment. Therefore, such cases largely seem to be ignored or missed, or they are just corrected during hypospadias repair without further mentioning them. This article aims at drawing the readers' attention to the association of penile torsion with hypospadias.

The method of measuring the degree of penile torsion is not clarified in most of the reports. In this study, we adopted the method of Sarkis and Sadasivam using a sterile small protractor for a better adjustment around the penile shaft [2]. In the literature, penile torsion was more commonly found in patients with anterior hypospadias (distal and mid penile), while it was absent in patients with posterior hypospadias [4,5]. In a more recent study carried out by Zeid and Soliman [5] torque was found to be associated with distal hypospadias in 32% cases of the cases, while none of the patients with proximal hypospadias had any torsion. In our study, the overall incidence of penile torsion associated with hypospadias was 31.6%. It was more common in association with distal (68.9%) than with proximal (10.34%) hypospadias.

The degree of torsion associated with hypospadias reported in the literature varies from 15° to 180° [4,5,10,11]; in our series the degree of torsion ranged from 15° to 110° with an average of 51.98° . Most studies in the literature [4,5,10,11] report a mild to moderate degree of torsion in the majority of cases. The same was found in our study, as severe torsion (>90°) was seen in only 11.2% of the cases.

In the majority of patients, the direction of congenital penile torsion is counterclockwise (to the left) [2,7]. In the present series, the right-to-left ratio was 1:3 with an incidence of right-sided torque of 29.31%. The reason for this higher incidence compared to that reported in the literature may be the fact that mild torsion was not taken into consideration in earlier studies.

An association between the severity of penile curvature and the severity of hypospadias has been reported [12] to be more common in proximal hypospadias. This complies with our findings. The mean chordee in proximal penile hypospadias was $73.58^{\circ} \pm 32.96^{\circ}$ compared to $38^{\circ} \pm 18.55^{\circ}$ in distal penile hypospadias (*P* value = 0.001). However, torsion was significantly more severe in distal hypospadias as compared to mid penile and proximal hypospadias, i.e. $62.38^{\circ} \pm 23.03^{\circ}$ vs. $38.04^{\circ} \pm 18.50^{\circ}$ and $18.25^{\circ} \pm 3.33^{\circ}$ (*P*=0.001).

Although we did not have the breakup of the incidence of torsion in distal, mid penile and proximal hypospadias among the total of 376 cases operated during the study period, this is probably the largest series reporting the various aspects of penile torsion in association with hypospadias and establishing the inverse occurrence of torsion and curvature in cases with hypospadias (Table 1).

Embryologically, at 8 weeks of gestation, the penile urethra is formed by fusion of the medial edges of the endodermal urethral folds, while the median raphe is formed by fusion of the ectodermal edges of the urethral groove. Mesodermal proliferation between these endo and ectodermal folds forms the corpora and other coverings of the penile shaft [13].

The pathogenesis of penile torsion lies in the eccentric fusion of the endodermal and/or ectodermal folds. This leads to misdirected mesodermal proliferation during formation of the corpora and, hence, aberrant attachment of the fascial coverings of the penis and spongiosum to one side, leading to torque [4]. Although various theories have been proposed to explain the occurrence of torsion, we believe torsion to occur due to abnormal attachments of the dartos fascia, Buck's fascia and skin. The median raphe forms by fusion of the ectodermal part of the urethral folds. Therefore, if during the development the fusion of endo and ectodermal components is eccentric (leading to torque), the whole penis rotates helically as a unit and the median raphe, being ventral, shifts to a direction opposite to that of the torque. Our findings are consistent with this theory as demonstrated in Fig. 2. However, in earlier studies, Sarkis and Sadasivam [2], Fisher and Park [3] and Zhou et al. [6] took the opposite view: they demonstrated that the median raphe deviates toward the side of the torque. The deviation is an important factor when torque is suspected in a newborn with unretracted skin, as it will then be a guide to assess the degree of torsion [1,2].

Ventral curvature was found in 61.2% of our patients, with the severity of curvature increasing the more proximal the meatus was located (38° in distal vs. 73.58° in proximal hypospadias). This is in keeping with the published literature [12].

Briefly touching upon the embryology of curvature, Kaplan and Lamm [14] who conducted studies on embryos suggested that during the development of the penis, ventral curvature with hypospadias may persist when normal development is arrested at an early stage. With this background, we propose that arrested development in hypospadias leads to a persistence of fetal ventral curvature. Since there is a proximal to distal fusion of the urethral folds, there is more time for the fetal curvature to straighten in distal hypospadias as compared to proximal hypospadias, thus explaining the increased severity of curvature in proximal hypospadias.

During this same stage of development, fusion of the endodermal and meso-ectodermal components takes place. When this fusion is eccentric, it leads to a torque which is evident from the eccentric median raphe. Since distal fusion takes place later than proximal fusion, distal fusion can proceed eccentrically for a longer time, thus causing more torque in distal hypospadias. In keeping with this hypothesis, torsion should be more common and more severe in distal hypospadias. In other words, the development of torsion is inversely proportional to the development of chordee.

Congenital penile torsion is a benign condition which may need no treatment, especially for mild degrees of torsion [14]. Isolated penile torsion correction is not necessary, if the degree of torsion is less than 60° because, apart from cosmetic disfigurement, the patients are asymptomatic. But if mild torsion is associated with hypospadias or chordee, the defect should be corrected along with hypospadias repair to achieve a cosmetically normal penis.

The technique of penile degloving and skin re-attachment suggested by Azmy and Eckstein [15] is effective only when repairing minor degrees of torsion [10,15]. The sutures would not be strong enough to hold the expanding and rotating force of erection in cases of moderate and severe torsion, which may lead to recurrence of torque. Furthermore, there is also a possibility of undercorrection which may also lead to recurrence of torque [11]. Resection of Buck's fascia and plication techniques are ineffective in severe torsion of

Type of hypospadias	No. of cases (116)	Range of torsion $15-110^{\circ}$ (mean $51.98^{\circ})^{a}$, range (mean \pm SD)	Correlation of severity of torsion with severity of hypospadias (total no. of torsion 100)			Range of chordee $15-125^{\circ}$ (mean 46.58°) ^b range(mean ± SD)	Correlation of severity of chordee with severity of hypospadias (Total no. of cases – 71; distal – 43, mid – 16 & proximal – 12)		
			Total no. of mild degree	Total no. of moderate degree	Total no. of severe degree		Total no. of mild degree	Total no. of moderate degree	Total no. of severe degree
Distal	80 (69%)	$25-110^{\circ}(62.38\pm23.03)$	28 (35%)	40 (50%)	12 (15%)	$15-90^{\circ}$ (38.00 ± 18.55)	32(74.42%)	09(20.93%)	02(4.65%)
Mid Penile	24 (20.65%)	$25-90^{\circ}$ (38.04 ± 18.50)	19(79.16%)	04(16.66%)	01 (4.16%)	$15-90^{\circ}$ (44.28 ± 21.11)	11(68.75%)	05 (31.25%)	00(0.00%)
Proximal Total	12(10.35%) 116	$1525^{\circ}~(18.25\pm3.33)$	12(100.00%) 59(50.86%)	00(0.00%) 44(37.94%)	00(0.00%) 13(11.20%)	$25-125^{\circ}$ (73.58 ± 32.96)	04 (33.33%) 47 (66.19%)	03 (25.00%) 17 (23.94%)	05 (41.66%) 07 (9.85%)

^a Correlation of severity of torsion with type of hypospadias:

Distal vs Mid penile: $X^2 = 55.39$, P = 0.0001. Distal vs proximal: $X^2 = 93.11$, P = 0.0001. Mid penile vs proximal: $X^2 = 11.98$, P = 0.001.

^bCorrelation of severity of chordee with type of hypospadias:-

Distal vs Mid penile: $X^2 = 25.40$, P = 0.0001. Distal vs proximal: $X^2 = 103$, P = 0.0001. Mid penile vs proximal: $X^2 = 39.95$, P = 0.0001.

Table 2Correlation of type of hypospadias with side of torsion (left or right).

Total no. of cases (116)	Cases of left sided torsion 82 (70.69%)	Correlation of severity of left sided torsion with severity of hypospadias			Cases of right sided torsion 34 (29.31%)		Correlation of severity of right sided torsion with severity of hypospadias		
		Total no. of mild degree	Total no. of moderate degree	Total no. of severe degree		Total no. of mild degree	Total no. of moderate degree	Total no. of severe degree	
Distal Penile (80)	61 (76.25%)	22 (36.06%)	30(49.18%)	09(14.75%)	19 (23.75%)	06(31.57%)	10(52.63%)	03(15.79%)	
Mid Penile (24)	15(62.50%)	13 (86.66%)	02(13.33%)	00(0.00%)	09(37.50%)	06(66.66%)	02(22.22%)	01(11.11%)	
Proximal (12)	06(50.00%)	06(100.00%)	00(0.00%)	00(0.00%)	06(50.00%)	06(100.00%)	00(0.00%)	00(0.00%)	
Total (116)	82 (70.69%)	41 (50%)	32(39.02%)	09(10.98%)	34 (29.31%)	18 (52.94%)	12 (35.30%)	04(11.76%)	

^aLeft side torsion varied from 15 to 110° (54.98).

^bRight side torsion varied from 15 to 95° (43.85)

^cCorrelation of severity of left side torsion with severity of hypospadias:

Distal vs Mid penile: $X^2 = 40.86$, P = 0.0001. Distal vs proximal: $X^2 = 96.29$, P = 0.0001. Mid penile vs proximal: $X^2 = 22.45$, P = 0.0001.

^dCorrelation of severity of right side torsion with severity of hypospadias:

Distal vs Mid penile: $X^2 = 2.09$, P = 0.350. Distal vs proximal: $X^2 = 46.48$, P = 0.0001. Mid penile vs proximal: $X^2 = 53.77$, P = 0.0001. more than 90° . Their disadvantages include a risk of recurrence if the sutures dissolve or give way early, some difficulty in achieving an exact correction and the possibility of producing counter torque as these techniques contradict the anatomical principles of surgery. In addition, these procedures entail significant operative risks and complications such as penile shortening and injury to the neurovascular bundle, leading to sexual dysfunction later [4,10,16]. The use of a dorsal dartos wrap [3] is an effective technique and feasible in hypospadias repair, however it is ineffective in cases of severe torsion as it carries the risk of recurrence, if the sutures give way. Other limitations of this technique are the difficulty of an exact correction of the torque and the possible necessity of an additional procedure for correction of chordee.

Urethral mobilization has been reported to be an effective method for the repair of torque and chordee [9]. The technique corrects the cause of torsion rather than producing a counter torque, as the etiological factors for both torsion and chordee are developmental deficits in penile facial coverings with the spongiosum and/or their attachment to the corpora and the glans. Mobilization of the spongiosum helps in performing tension-free suture spongioplasty which helps in preventing fistula formation and reconstructs a nearly normal urethra. Mobilization of the urethral plate and spongiosum from the meatus into the glans corrected torsion in 75% of our cases. Correction of torsion by penile degloving and skin re-attachment was feasible only in 11% of our cases. Mobilization of the urethra and corpus spongiosum releases the traction of fascial coverings and attachments of the spongiosum, and it corrects the torque. We have been able to correct penile torque in 89% cases by mobilizing the urethral plate and the divergent corpora spongiosa along with the proximal urethra and the urethral plate into the glans. The fact that correction of torque by penile degloving and mobilization of the urethral plate and spongiosum is feasible suggests that the attachment of dartos fascia, Buck's fascia and the urethral plate with the spongiosum to the corporeal bodies and glans may be the cause of torque, similar to chordee. However, further studies on a larger number of adult patients are still warranted.

Conclusions

Penile torsion with hypospadias is more common than reported. We should look for it while examining patients with hypospadias. The more distal the location of the hypospadiac meatus, the higher the degree of associated penile torsion. Ventral penile curvature is more common and more severe in proximal hypospadias cases. The deviation of the urethral meatus correlates well with the degree of torsion toward the side of the meatus, while the median raphe is attached to the opposite side. Penile torsion should be corrected along with hypospadias to achieve a cosmetically normal penis. The technique of mobilization of the divergent corpora spongiosa with the urethral plate into the glans corrects torsion in most cases. Hence, attachment of the urethral plate with the spongiosum to the corporeal bodies and the glans could be an etiological factor predisposing to penile torsion.

Conflict of interest

We have no conflict of interest to declare.

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