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Evaluation the outcome of two-stage repair in children with proximal hypospadias and severe chordee with hypospadias objective penile evaluation (HOPE) scoring

Proksimal hipospadiasli ve ciddi kordili çocukların iki aşamalı ameliyat sonuçlarının hipospadias objektive penil skorlama (HOPS) ile değerlendirilmesi

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Abstract

Introduction: Surgical management for the severe proximal hypospadias remains controversial and it might be challenging. In this study, we evaluated the surgical outcome of patients with proximal hypospadias managed by two staged repair with dorsal prepucial flaps.

Methods: Forty one boys with proximal hypospadias are enrolled between January 2006 and September 2016. The location of the urethral meatus was at the penoscrotal junction (PSH), at the scrotum (SH) or at the perineum (PRH). First stage; chordee was released and the prepucial flaps were constructed. Second stage; urethroplasty was performed according to Thiersch-Duplay principle. We assessed postoperative complication rates and utilized Hypospadias Objective Penile Evaluation (HOPE) scoring to evaluate the cosmetic outcome.

Results: There are a total of 41 boys (31 PSH (%76), 3 SH (%7) and 7 PRH (%17). The mean age at first operation was 15±5.3 months. The mean interval between the stages of the surgical procedures was 10.1±4.9 months. Sixteen patients had various enzyme deficiency or dysgenesis. In two patients, diverticulum is developed after the repair. stenosis has encountered in 10 patients and fistula in 5. Two patients had dehiscence at glanular level. Relation did not demonstrated between the severity of hypospadias and development of fistula, stricture and diverticulum (p>0.05). HOPE scores; 9.4 for the position of the meatus, 5.8 for shape of the meatus, 7.2 for shape of the glans penis, 9.3 for penile torsion and 9.8 penile curvature.

Discussion and Conclusion: Incorporating the dorsal prepucial skin flaps in staged procedures has both satisfactory cosmetic and functional success rates.

Keywords: Hypospadias objective penile evaluation (HOPE) grading; prepucial flaps; proximal hypospadias.

Özet

Amaç: Proksimal hipospadiasın cerrahi tedavisi halen tartışmalı olup bazen zorlu olabilir. Birçok teknik mevcuttur. Hangi tekniğin kullanılacağı cerrahın tercihine ve deneyimine dayanır. Biz bu çalışmada, proksimal hipospadiaslı çocuklarda uygulanan iki aşamalı prepusyal dorsal flap tekniğinin kozmetik sonuçlarını ve komplikasyon oranlarını değerlendirdik.

Gereç ve Yöntem: Ocak 2006 ile Eylül 2016 arasında proksimal hipospadias tanısı ile opere olmuş 41 çocuk çalışmaya alındı. Meanın konumuna göre üç grup oluşturuldu; penoskrotal seviye (PSH), skrotal seviye (SH), perineal seviye (PRH). Birinci aşamada kordi düzeltilmesi ve penis ön yüzünde prepusyal flep hazırlanması yapıldı. İkinci aşamada Thiersch-Duplay tekniğine göre üretroplasti uygulandı. Biz komplikasyonlarımızı ve hipospadias objektif penil skorlama (HOPS) kullanarak kozmetik sonuçları değerlendirdik.

Bulgular: Toplam hastaların, 31 tanesi PSH (%76), 3 tanesi SH (%7), 7 tanesi PRH (%17)'dir. Ortalama ilk operasyon yaşı 15±5.3 aydır. Aşamalar arasındaki ortalama süre 10.1±4.9 aydır. Onaltı hastada çeşitli enzim eksiklikleri veya disgenezi mevcuttur. İki hastada divertikül, 10 hastada stenosis ve 5 hastada fistül gelişmiştir. İki hastada glanüler seviyede dehisens mevcuttur. Hipospadias tiplerine göre bu komplikasyonlarda istatistiksel anlamlılık yoktur (p>0.05). HOPS skorlama sonuçları; meanın pozisyonu 9.4, meanın şekli 5.8, glansin şekli 7.2, penil torsiyon 9.3 ve penil kurvatür 9.8.

Sonuç: HOPS skorlama sonuçlarına göre iki aşamalı prepusyal flep tekniğinin kozmetik sonuçları kabul edilebilir seviyededir. Bu tekniğin sonuçları olumlu yönde etkilediği düşüncesindeyiz.

Anahtar Sözcükler: Hipospadias objektif penil skorlama (HOPS); prepusyal flepler; proksimal hipospadias.

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Proximal hypospadias with severe chordee comprises 20% of all hypospadias cases.[1] It is surgically challenging and there is no consensus for optimal surgical management of severe proximal hypospadias. There are four subgroups for proximal hypospadias; proximal penile hypospadias (PPH), penoscrotal hypospadias (PSH), scrotal hypospadias (SH) and perineal hypospadias (PRH).[2,3]

Various surgical techniques have been described so far. Although some surgeons manage with single stage with flap or graft, some others may prefer two stage with flap or grafts.[4–7]

Our aim is to evaluate the outcome of surgical correction of patients with severe hypospadias including complications and cosmetic outcome. They all are managed with two staged procedures by harvesting dorsal prepucial skin flaps. Only the severe proximal hypospadias cases were enrolled into the study as their management is the most challenging.

Materials and Method

Between January 2006 and September 2016, 41 boys were operated for severe proximal hypospadias. Thirty one patients had PSH. Three patients had SH and seven patients had PRH. We excluded all PPH mid shaft and distal hypospadias cases. All corrections are performed in two stages by using dorsal prepucial skin flaps. All surgical procedures (except orchidopexy if necessary) were performed after one year of age.

All patients had urethral chordee of more than 600. In all cases, urethral plate was transected to correct severe chordee during the first stage.

We utilized HOPE scoring to evaluate the cosmetic outcome (Table1).[8] HOPE scoring system evaluate the six surgical correctable topics including the position of meatus, shape of meatus, shape of glans, shape of penile skin, penile curvature and penile torsion. The total scoring range from 1 to the maximum 10 points.

Surgical Technique

First stage

The aim during the first stage is to provide a chordee-free penis and sufficient ventral penis and glanular groove for neourethra.

Glanular stitch for traction is placed. Circumferential circumcision incision 5-8 mm below the glans is performed. Urethral plate was transsected above the native meatus. Complete degloving of the penis is accomplished. The extent of transsection depends on the degree of urethral chordee and usually has a 5-10 mm distance to the native meatus. On the ventral side, all the structures leading to chordee are released. The grooves between cavernous and spongious bodies are isolated. An artificial erection test is performed to detect residual chordee, and dorsal midline Nesbit suture is performed if necessary. Afterwards glanular wings are seperated and mobilized to create enough space for glanular part of future urethra. In the next step, dorsal skin flaps are harvested. Dorsal preputium is incised on the midline vertically and transposed ventrally. Both wings of the flaps are sutured beneath the glanular wings deeply and fixed to the Buck's fascia ventrally with Polydioxanone (PDS)® sutures and polypropilen 2/0 or 3/0 is preferred for Nesbit plication when necessary. Silicon catheter is placed into bladder and pressure dressing is applied to prevent hematoma formation. The bladder catheter is removed after 4 days.

Second stage

Main goals of the second stage are; construction of urethral tube with sufficient calibration (tubularization stage), and satisfactory cosmetic appearance of glans and urethral meatus.

Neo-urethra is constructed with previously incorporated prepucial skin flaps that has adequate vascularization. Urethroplasty is performed according to Thiersch-Duplay principle. When calibration of neourethra is deemed insufficient, uretral plate is incised in the midline at the dorsal aspect of the neourethra as in the Snodgrass procedure. We did not use penile tourniquet for hemostasis in any case. Un-interrupted suturing with PDS® 6/0 is used for urethroplasty and interrupted second layer interrupted stiches with PDS® 7/0 performed to reduce the tension on the anastomosis. Thereafter, neo-urethra is covered with well-vascularised adjacent subcutaneous tissues with interrupted PDS® 6/0 sutures. Bladder is drained with an appropriate sized urethral catheter that usually stays more than 10-12 days. Pressure dressing is routinely applied.

For the statistical analysis descriptive statistic Chi-square test, Fisher exact test, ANOVA were used.

Results

There were forty one boys with severe hypospadias; 31 penoscrotal (75,6%), 3 scrotal (7,3%) and 7 perineal (17,1%) (Table 2).

All surgical procedures (except orchiopexy) were performed after one years of age. The mean age at first operation is 15±5,3 months old. The mean interval between the stages of the surgical procedures is 10.1±4,9 months (7.6-14.5).

Sixteen patients had diagnosed as partial androgen insensitivity syndrome (PAIS) (39,02%), 2 as 5 α-reductase deficiency (4,8%), one as mixed gonadal dysgenesis (2,4%), and one as 3-beta hydroxysteroid dehidrogenase deficiency (Table 3).

In all cases, degree of urethral chordee was more than 600. Postoperative chordee is less than 20 0 in all patients. Nesbit sutures are performed in 12 patients after urethral plate transsection (29,2%). Midline vertical incision of urethral plate for tension-free anastomosis was not necessary in most of the patients (85,4%). We performed urethral plate incision as in the Snodgrass procedure only in 6 patients.

Before first stage of operation, androgen ointment was prescribed to increase the size of penis in 24 patients (58.5%) (total 50 sessions; three cycles in 7 patients, 2 cycles in 12 patients, single cycle in 5 patients).

Urethral diverticulum is developed in 2 patients after the second stage of the procedure (4,8%) at the proximal penile and

Table 1. Hypospadias objective penile evaluation (HOPE)

Position of meatus	Position 1 (10 points) Glanuler	Position 2 (8 points) Subcoronal	Position 3 (5 points) Coronal	Position 4 (3 points) Midshaft	Position 5 (1 point) Proximal
Shape of meatus	Normal (10 points)	Slightly abnormal (7 points)	Moderately abnormal (4 points)	Severely abnormal (1 point)	
Shape of glans	Normal (10 points)	Slightly abnormal (7 points)	Moderately abnormal (4 points)	Severely abnormal (1 point)	
Shape of penil skin	Normal (10 points)	Slightly abnormal (7 points)	Moderately abnormal (4 points)	Severely abnormal (1 point)	
Torsion of penis	0^0–30^0 (10 points)	30^0–50^0 (7 points)	50^0–70^0 (4 points)	$>70^0$ (1 point)	
Curvature in penile erection	0^0–30^0 (10 points)	30^0–50^0 (7 points)	50^0–70^0 (4 points)	$>70^0$ (1 point)	

Table 2. Original meatal location before correction

	Number (n)
Penoscrotal (PSH)	39
Scrotal (SH)	8
Perineal (PRH)	10

Table 3. Number of related syndromes

	Number (n)
Partial androgen insensitvity syndrome (PAIS)	21
5 α-reductase enzyme defiency	4
Mixed gonadal dysgenesis	2
3-beta hydroxysteroid dehidrogenase deficiency	1

Table 4. Number of complications

	Number (n)
Urethral diverticulum	3 (2 scrotal, 1 proximal penile)
Meatal stricture	12
Urethral fistula	7 (3 subcoronal, 2 coronal, 1 midshaft, 1 penoscrotal)
Dehiscence	2 (subcoronal)

scrotal level, respectively. Both are corrected surgically. Stricture of the urethral meatus developed in 10 patients (24,3%). They received meatotomy and urethral dilatations (a total of 18 dilatations). Urethral fistula developed in 5 patients (12,1%) (2 subcoronal, 2 coronal, 1 midshaft fistula). All fistulas are corrected in one stage except one which is corrected in two stage. Number of patients with dehiscence at subcoronal level was 2 (4.8%). We performed MAGPI and Mathieu procedures for these patients afterall (Table 4). Eight patients had scrotal transposition (19.5%). Transpositon of the scrotum was corrected at older ages but before 5 years of age in all the patients.

Table 5. Mean HOPE Scores of the study group. As all the patients underwent circumcision for religious reasons, scoring for prepucium is not performed

Position of meatus	9.5
Shape of meatus	6.2
Shape of glans	7.7
Torsion of penis	9.4
Curvature in penile erection	9.7

Incidence of urethral fistula is significantly lower in PSH than PRH group (p=0,002). No statistical significance is demonstrated between fistula (0.41), stricture (0.53) or diverticulum (0.74) with the severity of hypospadias (p>0,05). Neither interval between the stages of correction (p=0.57), nor the age at the first stage (p=0,43) is found to be related with uretral stenosis. Age at the first stage (p=0,47), interval between the two stages (p=0,32) are not found to have an impact on fistula development. No significant relation is demonstrated between preoperative local testosterone treatment and fistula or stenosis formation (p=0,665 and p=0,606, respectively). Fistula is observed significantly higher in patients with transposition of scrotum (p=0,039). Thirty two patients (78,04%) had undescended testis (11 bilateral and 16 non-palpable).

When HOPE scoring system is utilized to evaluate the cosmetic outcome, mean scores were as follows; 9,4 for position of the meatus,5,8 for shape of the meatus, 7,2 for shape of the glans penis is, 9,3 for torsion and 9,8 for penile curvature (Table 5).

Disscussion

Hypospadias is characterised with deficient ventral urethral structures of the penis.[9,10] The management depends on location of meatus, size of penis and glans, width and depth (groove) of urethral plate, severity of chordee and surgeon's experience.[5,10] The main goals in hypospadias surgery are to reconstruct straight penis and slit like meatus on the tip of glans.[11]

Proximal hypospadias constitute about 20% of all hypospadias cases. There are mainly four types of proximal hypospadias; proximal penile (PPH), penoscrotal (PSH), scrotal (SH) and perineal (PRH).[12] In PPH, chordee may be corrected without transection of urethral plate and can be managed with single staged procedures. Therefore, PPH cases who did not require urethral transection are excluded from this study. In our opinion, the main cause of chordee is straining due to urethral plate in proximal hypospadias including PSH, SH, and PRH. Therefore, transection of urethral plate is inevitable in these cases for straight penis and sufficient cosmetic outcome.

In literature, several procedures for the treatment of severe proximal hypospadias have been described. Some surgeons prefer single stage surgery after transection of urethral plate by using flap or free grafts.[5,13–17] On the other hand, many others suggest that two staged procedures (grafts, flaps) provides better cosmetic and functional results.[18–21] For the construction of neourethra, flaps and grafts are used and all have certain complications. One of the important factor to prevent complications such as fistula, stenosis, diverticula is well vascularization of flaps or grafts, whatever is used. In proximal hypospadias, usually a flat and narrow urethral plate is dealed with and it must be augmented for adequate tubularization. The quality of urethral plate is an important factor for surgical success. We think that urethral augmentation and vascularization of flaps are satisfactory in two stage management with prepucial flaps. One more benefit of the flaps are providing enough tissue to cover the urethroplasty with a second layer harvested from adjacent subcutaneous tissues. This coverage is deemed critical to prevent fistula formation. Our main concern is to provide tension-free anastomosis in most of our patients and we did not find necessary to incise dorsal urethra in midline in most of our patients (85,4%).

Our postoperative incidence complications rates including meatal stricture, fistula, diverticulum and dehiscence are acceptable level. HOPE scoring is defined by 'Dutch hypospadias study group' which evaluates penile appearance by following 6 items; the position of meatus, shape of meatus, shape of glans, shape of penile skin (exculede from our study) and penile torsion (8). The penis is photographed by 5 different views and assessment by pediatric urologists. Another scoring system; Hypospadias objective scoring evaluation (HOSE) was described by Holland et al (22) before HOPE. In HOSE scoring,the 5 items were evaluated including the meatal location, meatal shape, urinary stream, erection and fistula. HOPE scoring is more actual and more comprehensive than HOSE scoring so we used HOPE scoring in our study. HOPE scores are high especially for position of meatus, and curvature of penis in our study. Relatively lower scores for the shape of glans and meatus might be attributed to the inherited narrow structure of the glans and shallow nature of glanuler urethral plate in these severe cases of hypospadias.

Cosmetic outcome of two staged correction of severe hypospadias by using dorsal prepucial skin flaps is successful according to HOPE grading system and complication rates are reasonable. In our opinion, incorporating the dorsal prepucial skin flaps in staged procedures has both satisfactory cosmetic and functional success rates.

Conflict of interest: There are no relevant conflicts of interest to disclose.

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