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Original Research Article

Hypospadias with cryptorchidism presented as ambiguous genitalia and its management with surgery

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ABSTRACT

Background: The incidence of hypospadias with cryptorchidism is about 6-31.6%. Current management recommendations are that undescended testis beyond three months needs surgery between 6-12 months of age. Proximal hypospadias and cryptorchidism overlap with disorders of sex development and endocrine problem. The aim of this study was to assess the outcome of surgical management of hypospadias with cryptorchidism, at selected centers in Dhaka, Bangladesh.
Methods: This prospective study was conducted in the Department of Pediatric Surgery of Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh during the period from January, 2018 to January, 2022. In this study, we included 70 diagnosed cases of hypospadias with cryptorchidism presented as ambiguous genitalia who underwent surgery.
Results: Mean age at surgery was 4.6±1.4 years and majority (91.4%) were male. Associated renal anomalies were absent in 81.4% cases, symptomatic renal anomalies were present in 11% cases, severe chordee was 62.9%. Abnormal hemiscrotum was 98.6% and normal hemiscrotum was 1.4%. Hernia with cryptorchidism was present in 30 % cases. Testosterone was normal in 94.3% patients, estrogen and progesterone was elevated in 5.7% patients. After operation, male was 94.3% and female was 2.9%, chordee condition was corrected in 81.4% and urine passed through tip of penis in 84.3% patients.
Conclusions: We found that surgery is simple, safe, and effective in the treatment of patients with hypospadias and cryptorchidism with satisfactory functional results, normal physical health status, improved quality of life, satisfying psychological support and probably less pain.

Key words: Hypospadias, Cryptorchidism, Pediatric surgery

INTRODUCTION

Undescended testis (UDT), also known as cryptorchidism, and hypospadias are considered as two common urogenital malformations in young boys.[1] The incidence of hypospadias and concomitant UDT is about 6% and 31.6% respectively.[2] Hypospadias, a condition where the urethra opens on the underside of the penis with associated ventral penile curvature, is the second most common genital birth defect in boys, following cryptorchidism. With an incidence of one in 200 live male births, hypospadias correction is one of the common surgical procedures performed by pediatric urologists.[3,4] Hypospadias is the second most frequent congenital condition in boys after

cryptorchidism, with an incidence of 0.3-0.7% compared to 2-4% for cryptorchidism.[5]

Surgery should be offered for all forms of hypospadias, but there is no consensus on the minimum age for the surgery. The results of this surgery pose a problem because of its many complications. Since the 1980s, single-stage operations, such as the one described by DUCKETT, have been adopted by some authors.[6,7] Surgery for hypospadias can be done in one or more operative stages. Each technique has its own advantages, but one-stage procedures are often associated with complications and require reoperations.[8] The complication rates were 20%-50%.[8] The debate over the optimal treatment for hypospadias is ongoing and most believe that a two-stage procedure offers superior functional and cosmetic results with fewer complications like the Bracka technique. These procedures seem to be more adapted in our developing countries where the cost of surgery remains inaccessible to the majority of the patients. A few studies have evaluated the results of DUCKETT's procedure for hypospadias in our countries.[9] The occurrence of hypospadias and concomitant UDT needs early surgical intervention. It should be highlighted that when a patient is diagnosed with hypospadias and concomitant UDT, simultaneous intervention for treatment of these two anomalies would be of great value to prevent multiple surgeries. Additionally, application of a technique with satisfactory cosmetic results, normal appearance, and less postoperative discomfort can be beneficial in patients with concomitant hypospadias and UDT. The surgical treatment of UDT should be instituted as early as possible due to the fact that the preservation of spermatogenesis is commonly lost after puberty and the occurrence of spermatic cord torsion and strangulated hernia may increase.[1,10] Reconstruction of the urethra and penis with satisfactory cosmetic and functional outcomes, the least complication rate and reduction in number of surgeries are among the aims of hypospadias repair. Several surgical techniques are presented for hypospadias repair among which the tabularised incised plate (TIP) urethroplasty which is a single stage procedure, has been considered as a technique of choice especially in proximal types.[11] A person's sexual orientation can be determined by a variety of factors, such as their chromosomal pattern (XY OR XX), type of gonads (ovary or testis), dominant sex hormone (estrogen or androgen), topography of their genitalia, and secondary sexual features. In essence, chromosomal structure is genetically programmed for gonad nature, which denotes the presence of circulating sex hormones that influence phenotypic and genital characteristics. A child with hypospadias and cryptorchidism is extremely difficult for a pediatric surgeon to manage. Hypospadias with cryptorchidism is a social emergency. Therefore, in this paper, we aimed to assess the outcome of surgical management of proximal penile hypospadias with cryptorchidism.

Objectives

The main objective of the study was to assess the outcome of surgical management of proximal penile hypospadias with cryptorchidism, at selected centers in Dhaka, Bangladesh.

METHODS

This was a prospective study and was conducted in the department of pediatric surgery of Bangabandhu Sheikh Mujib medical university (BSMMU), Dhaka, Bangladesh during the period from January, 2018 to January, 2022. In this study, we included 70 diagnosed cases of hypospadias with cryptorchidism presented as ambiguous genitalia who underwent surgery. These are the following criteria to be eligible for enrolment as our study participants: Patients aged up to18 years; Patients diagnosed case of hypospadias with cryptorchidism; Patients with undescended testis admitted in Neonatology and Paediatric Surgery Department; Patients who were willing to participate were included in the study and Patients diagnosed case of hypospadias without UDT, Patients with distal penile hypospadias; Patients diagnosed case of cryptorchidism without hypospadias; Patients with known allergy to anesthetic drugs; Patients with any history acute illness (e.g., renal or pancreatic diseases, ischemic heart disease etc.) were excluded from our study.

Surgical procedure

Reconstructive surgery was done specially Genitoplasty where required with team decision including patient party. Feminizing genitoplasty: cliteroplasty, vaginoplasty, labiaplasty, orchiectomy, penectomy-according to need. Masculinizing genitoplasty: Hysterectomy and bilateral salpingo-oophorectomy, Mastectomy Orchidopexy. Orthoplasty, Urethroplasty-according to need.

Statistical analysis

All data were recorded systematically in preformed data collection form and quantitative data was expressed as mean and standard deviation and qualitative data was expressed as frequency distribution and percentage. Statistical analysis was performed by using SPSS (Statistical Package for Social Sciences) for windows version 10. Probability value <0.05 was considered as level of significance.

RESULTS

In this study, we included 70 patients and mean age at surgery was 4.6±1.4 years (Table 1). Sex distribution at diagnosis, 91.4% were male and 8.6% were female (Table 2). Associated renal anomalies was absent in 81.4% cases and symptomatic renal anomalies was present in 11% cases and only 2.7% were symptomatic and 11.7% were asymptomatic.

Table 1: Age distribution of our study population.

Age (years)	N	%	P value
0-9 years	45	64.28	<0.0001
10-18 years	25	35.71	
Mean±SD (years)	4.60±1.48		

Table 2: Gender distribution of our study patients after Karyotyping.

Gender	N	%
Male	64	91.4
Female	6	8.6
Total	70	100.0

Table 3: Distribution of our patients by associated renal anomaly and urinary continence.

Renal anomaly		N	%
Valid	Symptomatic	2	2.9
	Asymptomatic	11	15.7
	Absent	57	81.4
	Total	70	100.0
Urinary continence	Incontinence	5	7.1
	Continence	65	92.9
	Total	70	100.0

Table 4: Distribution of our study patients by degree of chordee.

Parameters		N	%
Valid	Moderate	26	37.1
	Severe	44	62.9
	Total	70	100.0

Table 5: Development of hemiscrotum.

Parameters		N	%
Valid	Normal	1	1.4
	Abnormal	69	98.6
	Total	70	100.0

Table 6: Distribution of our study patients by the condition of the testis.

Parameters	N	%
Palpable	17	24.3
Ectopic	1	1.4
Non-Palpable	52	74.3
Total	70	100.0

In the present study urinary continence was present in 93% and incontinence was present in 7% cases (Table 3). In this study we found severe chordee was 37% and moderate and mild type was 55.7% and 7.1 % respectively (Table 4). We found abnormal hemiscrotum was 10% and under development of hemiscrotum was 88.6% and normal hemiscrotum was only 1.4% in this study (Table 5). We observed that testis was nonpalpable in 74.1% cases and ectopic testis was 1.4% and palpable testis was 14.1% (Table 6).

Table 7: Distribution of our study patients by associated hernia with cryptorchidism and hormone analysis.

Hernia with cryptorchidism		N	%
Valid	Present	21	30.0
	Absent	49	70.0
	Total	70	100.0
Hormone analysis	Testosterone normal	66	94.3
	Estrogen and progesterone elevated	4	5.7
	Total	70	100.0

Table 8: Distribution of our study patients by operation performance and stages.

Operation performance	N	%
Staged procedure	67	95.7
Single time	3	4.3
Total	70	100.0
Stage 1		
Done	70	100.0
Stage 2		
Done	67	95.7
Not done	3	4.3
Stage 3		
Done	25	35.7
Not done	45	64.3

Table 9: Distribution of study population by outlooking of, condition of chordee, state of penile erection, urine passes through tip of penis after the operation.

Genital organs	N	%
Male	66	94.3
Female	2	2.9
Chordee condition		
Corrected	57	81.4
Residual	13	18.6
Penile erection		
Normal	54	77.1
Abnormal	16	22.9
Urine passes through tip of penis		
Yes	59	84.3
No	11	15.7

Among all patients associated hernia with cryptorchidism was present in 30% cases. Among all patients, testosterone was elevated in 94.3% of patients, and estrogen and progesterone were elevated in 5.7% of patients (Table 7). Staged procedure was done in 95.7 % and single time was done in 4.3% patients. In our study stage 1 was done in

100% patients, stage 2 was done in 95.7% and stage 3 was done in 35.7% patients (Table 8). After operation, male was 94.3% and female was 2.9%, chordee condition was corrected in 81.4% and residual in 18.6%. We found penile erection was normal in 77.1% and urine passed through tip of penis in 84.3% patients (Table 9).

DISCUSSION

The standard treatment for hypospadias is surgery. It is a tough procedure with many techniques outlined. The most sophisticated methods, with a higher risk of complications, are required for the posterior variants of hypospadias.[12] Some authors use two-stage approaches to improve their outcomes.[7,13] These might lower the risk of infection and failure caused by fap necrosis or suture disunion.[7] Zheng et al advocate two-stage urethroplasty in posterior forms with considerable curvature following urethral plate removal to avoid the frequent urethral stenosis seen with single-stage urethroplasty.[13] Bankole Sapin et al discovered a 26% complication rate for two-stage surgery and a 37% complication rate for one-stage surgery.[7] The Bracka technique is the most successful in this indication.[14] The choice of the DUCKETT's one-step technique was guided by the need to excise the urethral plate as part of the urethroplasty, the presence of a posterior preputial apron, but also because of the good mastery of the technique.[6] According to some writers, single stage urethroplasty produces good results, particularly in young patients.[15] Others, however, such as Bankole et al consider that difficulties are more frequent.[7,13] Hypospadias surgery complications are more common in underdeveloped countries.[7,16] Other authors, particularly in Western countries, have reported lower complication rates.[12,17] Wound infection, fap necrosis, urethra-cutaneous fistula, and neo-urethral or neo-meatus stenosis are the most common consequences of hypospadias surgery.[16-19] Wound infection was the most common early complication in this series, which was most likely attributable to the poor post-operative care circumstances, but also to the hot and dry tropical climate. The use of two-stage surgery could in some cases minimize this complication rate. These infections are promoted to the failure of the repair by the sutures being loosened or by fap necrosis.[20] Flap necrosis can also be the consequence of poor dissection and devascularisation of the fap. Diallo et al reported 15% fap necrosis and suture disunion.[10] Urethral stenosis is usually the result of devascularization of the stenotic area in the case of tension anastomosis, with a higher risk of wound infection.[16] The urethral stenosis rate is variable according to the literature. The rate was high in the series of Akbiyik et al who reported a 27.5% stenosis rate, while Mosharafa et al reported only 0.9% urethral meatus stenosis. Finally, the rate of unintrusive residual penile curvature (5%) was lower than that reported by Diallo et al.[12,16,18] In total, the success rate of the DUCKETT technique was 63% after treatment of complications. The rate of good results depends, among other things, on the technique, the operator's experience, the type of hypospadias, and the presence and severity of the curvature.[22] Diallo et al in Guinea, after a 3-22 month follow-up, reported 61.7% good results for all techniques and all forms of hypospadias.[16] While Diao et al in Senegal reported 62.5% good results for all previous forms of hypospadias, all techniques combined.[19] Other, mainly Western authors such as Dewan et al report better success rates with the Duckett technique after a single treatment (66.6% success out of 190 hypospadias treated). Similarly, Zheng et al reported only 24% complications after a first treatment with a pedicled preputial fap.[21,23]

Cryptorchidism is the most common malformation of male sexual development, with an overall prevalence rate of 3% in full-term newborn males. Most palpable UDT spontaneously descends within the first months of life, decreasing the prevalence to 1-2% for boys aged 6 months to one year.[24-27] The right side is affected more often than the left, occurring in 70% of cases. The majority of cases present with a palpable UDT, non-palpable testes accounting for approximately 20% of all cases.[25] This tendency was similarly reflected in the patient population of this study where no palpable testes were found in 74.3% of cases. The undescended testis is a risk factor for both infertility and testicular malignancy. The histopathological and functional changes of the cryptorchid testis are well established with defective spermatogenesis and the loss of endocrine function being early consequences.[25,26,28-31] The histopathological hallmarks with degeneration of Sertoli cells, decreased numbers, and defective maturation of germ cells, as well as hypoplasia of the Leydig cells, are evident from as early as one year of age.[25,28,32,33] Bilateral UDT is more significantly correlated with infertility, with azoo- or oligospermia reported in 66-87.5% of cases, compared to 33-35% of cases with unilateral UDT.[27,28, 32,34] Normal fertility rates are reported in 87-90% of patients undergoing repair within the first 2 years of life.[27,28,32,35,36] This figure is similar to fertility levels found in the general population.[35] Abnormal spermatogenesis is reported to be as high as 98% in the cryptorchid testis of post-pubertal men.[16] Fertility assessment was not part of the routine investigative protocol in this cohort; however, the relevance is noted by the above percentages and should be included in future studies in refining management guidelines. Besides the malignant potential and the loss of function of the cryptorchid testis influencing the approach to the management of UDT, other factors that have been recorded as fundamental are the apparent increased risk of the UDT for undergoing torsion, the abnormal position predisposing it to trauma and the psychological benefit of having intrascrotal testes.[27] Early surgical correction is still the gold standard of the management of UDT.[24,27,32,36-39] Guidelines for pre-pubertal patients are well-defined; however, those applicable to the post-pubertal patient seem far more rudimentary and have remained unchanged for a number of years. To the best of our knowledge no series has addressed or identified the challenging factors and evaluated the outcome of management of hypospadias with cryptorchidism child. In this study we aimed to do that. In our study mean age at surgery was 4.6±1.4 years,

91.4% were male and 8.6% were female. Associated renal anomalies was absent in 81.4% cases and symptomatic renal anomalies were present in 11% cases and only 2.7% were symptomatic and 11.7% were asymptomatic. We found severe chordee was 62.9% and moderate type was 37.1%. We found abnormal and under development of hemiscrotum was 98.6% and normal hemiscrotum was only 1.4% in this study. Testis was non palpable in 74.1% cases and ectopic testis was 1.4% and palpable testis was 14.1%. Among all patients associated hernia with cryptorchidism was present in 30 % cases. The mean length at first visit was 37.62±10.52 cm. Among all patients, testosterone was normal in 94.3% of patients, and estrogen and progesterone was elevated in 5.7% of patients. Staged procedure was done in 95.7 % and single time was done in 4.3% patients. After operation, male was 94.3% and female was 2.9%, chordee condition was corrected in 81.4%, penile erection was normal in 77.1% and urine passed through tip of penis in 84.3% patients.

Limitations

Small sample size was a major limitation due to short study period. There are more complications or adverse effects of surgical management of hypospadias with cryptorchidism child needs to be evaluated. After evaluating those patients we did not follow up with them for a long term and have not known other possible interference that may happen in the long term with these patients.

CONCLUSION

In our study, we found that surgery is the only treatment in the management of patients with hypospadias and cryptorchidism presented as ambiguous genitalia. The findings of the current study demonstrated that surgery is simple, safe, and effective in the treatment of patients with hypospadias and cryptorchidism with satisfactory health status, improved quality of life, a cosmetically appealing acceptable. So further study with a prospective and longitudinal study design including larger sample size needs to be done for sex differentiation and acceptable genital organ.

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