

CASE REPORT

Urethral stricture in adult as a consequence of childhood hypospadias repair

Moniruzzaman M¹, Faruquzzaman², Mohiuddin T³

Union health sub-centre, Barakpur, Khulna, Bangladesh. drfaruquzzaman@yahoo.com

Abstract: We report our experience in treating such patients. A retrospective chart review was performed from 2004 through 2007 in Urology Department of Dhaka Medical College Hospital by using the convenient type of purposive sampling. In cases undergoing perineal urethrostomy, we suggested the “side-to-side” technique, which comprised longitudinal urethrotomy and everting the mucosal and submucosal layers of the urethra to the incised skin. Graft-based urethroplasty using buccal mucosal graft, in one-stage or two-stage repairs, was the preferred choice of formal reconstruction. Patients undergoing hypospadias surgery should receive lifelong follow-up protocol to detect latent urethral strictures (Fig. 8, Ref. 12). Full Text in free PDF www.bmj.sk.
Key words: urethral stricture in adult, childhood hypospadias repair, retrograde urethrography.

Hypospadias is a common congenital abnormality occurring in 1/300 live births, and is the most common congenital penile anomaly (1, 2). Numerous surgical techniques have been developed to correct this anomaly. However, no single method is considered the standard of care, and they all share the common complications of occasional urethrocutaneous fistula and urethral strictures. The incidence of urethral stricture after hypospadias surgery in pediatric population is reported, and occurs in about 6.5 % after short follow up (3). On the other hand, there are few reports dealing with urethral strictures in adults after they had hypospadias surgery in childhood. In the current series, we described our experiences in 9 such cases, and review their particular characteristics and suggested treatments.

On the Figure 1 is an example of an infant with imperforate anus. Note the anal dimple and in addition there is a hypospadias involving the glans. Anal atresia is often associated with other anomalies, primarily genitourinary tract or vertebral anomalies. It is also observed in the VACTERL syndrome(R).

Figure 2 shows this infant with a bifid scrotum which indicates failure of the testosterone-induced fusion of the two



Fig. 1. An infant with imperforate anus.



Fig. 2. A bifid scrotum with indicates failure of the testosterone-induced fusion of the two labioscrotal folds.

¹Union health sub-centre, Barakpur, Khulna, Bangladesh, ²Chittagong Medical College Hospital Chittagong, Bangladesh, and ³Sher-e-Bangla Medical College Hospital Barisal, Bangladesh

Address for correspondence: Faruquzzaman, Dr, C/O: Md. Moazzem Hossain (Engineer), House No. 7, Islambag Road (Paulpara), Railygate, Daulatpur (Postal code 9202), Bangladesh.
Phone: +8801726124840

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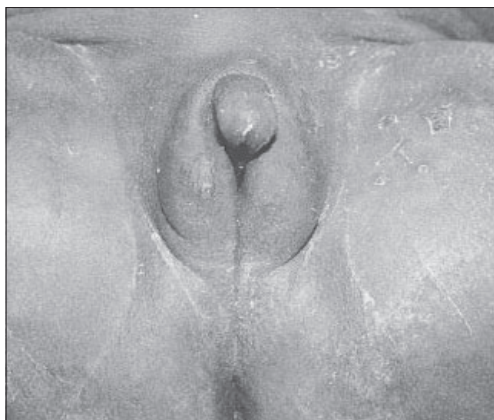


Fig. 3. Ambiguous genitalia.



Fig. 4. Genital adrenal hyperplasia in a female infant.

labioscrotal folds at about 10 to 11 weeks of gestation with partial penoscrotal transposition. The scrotum is divided into two halves, each containing a testis; this condition may be associated with other perineal abnormalities such as hypospadias or imperforate anus (R).

Figure 3 shows a female infant with ambiguous genitalia. The karyotype was XX. Difficulties in determining the sex of an infant may arise from abnormalities of the external genitalia. Ambiguous genitalia encompass a wide range of abnormalities having their origin before the 12th week of gestation. The phallus commonly shows hypospadias with chordee formation and appears large in proportion to the persisting labioscrotal folds which may or may not contain gonads (testis, ovotestis, or rarely a well-defined ovary). In females, the labia may be fused and the clitoris hypertrophied. This condition is known as pseudohermaphroditism (R).

Another example of congenital adrenal hyperplasia in a female infant (Fig. 4). Note the marked labioscrotal folds; testes were absent, and a hypospadias is present. In 21-hydroxylase deficiency, genitalia are conspicuously abnormal at birth. The degree of masculinization can be judged by the size of the clitoris and the degree of labioscrotal fusion, which determines the size of the urogenital sinus. The phallus is invariably enlarged,

often approximating the size of a penis. It is generally bound with chordee, behind which a perineal hypospadias is situated. Commonly the labia majora have the appearance of a bifid scrotum. The orifices of the vagina and the urethra lie within the perineal opening of the urogenital sinus(R).

Methods and materials

Type of study: A retrospective study.

Place of study: Urology Department of Chittagong Medical College Hospital.

Type of sampling: Convenient type of purposive sampling.

Methods

A retrospective chart review was performed from 2006 through 2008. Nine consecutive adult patients who had current urethral strictures and had undergone childhood hypospadias surgeries were included in this study. All adult urethral strictures were managed by a single surgeon. The strictures in these patients were all symptomatic and were documented by retrograde urethrography. Information regarding hypospadias repairs, previous urethral manipulations, presenting symptoms, stricture length, definite treatment, and short-term outcomes were obtained from medical records. In cases undergoing perineal urethrostomy, we suggested the "side-to-side" technique, which comprised of longitudinal urethrotomy and everting the mucosal and submucosal layers of the urethra to the incised skin. Graft-based urethroplasty using buccal mucosal graft, in one-stage or two-stage repairs, was the preferred choice of formal reconstruction.

Data processing and analysis

Using computer based statistical package statistical analysis of the data was done. Data was analyzed with SPSS computer package programme. The survey data was usually analyzed using descriptive statistics, such as mean, SD, percentage, coefficient of variation etc. All parameters were expressed as mean \pm standard error of the mean (SEM).

Report was produced by computer based programme Microsoft Word, Power point, Adobe and other accessories. A p value of less than 0.05 was considered as significant.

Results

Mean patient age was 38.6 years, and mean follow up period was 1.9 years. All patients had their primary 2 hypospadias surgeries between 1 and 12 years old. The lag time of the adult urethral stricture presentation ranged from 25 to 57 years, with an average of 36 years between hypospadias surgery and presentation to our clinic with urethral stricture. Four of the 9 patients (44 %) presented with acute urinary retention, and one of these patients developed acute renal failure due to prolonged urinary retention, before stricture was diagnosed. The other associated complications included fistula in one case. Only three of the 9 patients (33 %) suffered from lower urinary tract symptoms (LUTS) including decreased voiding stream, spraying, drib-



Fig. 5. Typical complex urethral stricture after childhood hypospadias repair, with a distal penile location and complicating fistulae.

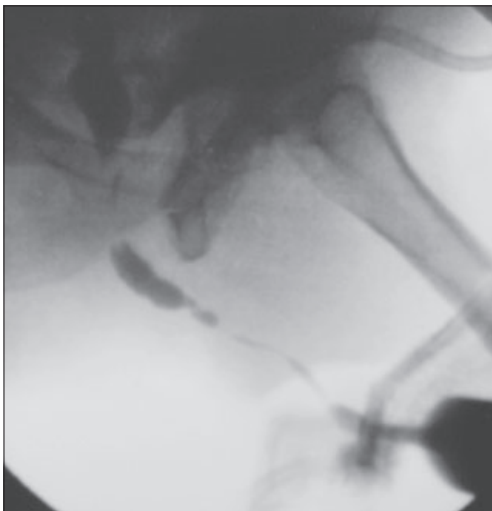


Fig. 6. Typical retrograde urethrogram appearance of adult stricture after hypospadias repair showing a long stricture sparing only the bulbar urethra.

bling, and nocturia. In one case, urethral stricture was discovered when we evaluated unresolved urethrocutaneous fistulas (Fig. 5). Before transferred to our institute, 5/9 patients had undergone endoscopic treatment for strictures, and 2/9 had failed open urethroplasties.

Penile urethra was involved in all cases, and bulbar urethra was involved in 5/9. The stricture length ranged from 1 to 17cm (mean, 10.3cm) (Fig. 6). Open urethroplasties with buccal mucosal grafts were performed in 4/9 cases: two with single-stage repair and two with 2-stage repair with buccal grafts (Fig. 7). Salvage perineal urethrostomy was performed in 2/9 cases, usu-



Fig. 7. First-stage urethroplasty with buccal grafts.



Fig. 8. Perineal urethrostomy performed using our suggested "side-to-side" technique.

ally in patients who did not wish complex definitive urethral surgery (Fig. 8). The perineal urethrostomy was planned to be permanent. Another 3 cases chose to receive repeated endoscopic treatments (direct visual internal urethrotomy (DVIU) and/or dilatations) although perineal urethrostomy or urethroplasty had been offered. None was cured by DVIU/dilatations. Four patients receiving open urethroplasty with buccal grafts were free of stricture recurrence. Stenosis of urethrostomy developed in one case and was successfully managed by a V-Y plastic technique.

There is little literature mentioning adult urethral stricture in hypospadias patients. Barbagli et al. published a series of 60 adults with previously failed hypospadias repair (4), including 34 cases that underwent treatment for urethral strictures. Their

overall successful rate was 75 % (83 % for one-stage repair, 68 % for multistage repairs). It was evident that those who needed multistage repair plans were at higher risk of failure because they had more severe strictures and extremely poor quality native tissue than those in whom the surgeons would consider risking a single-stage repair.

While differences in wound healing ability between children and adults are well described (5), little direct data is available on the relative behavior of adult and childhood tissue in the urologic arena. Adult hypospadias surgery has been reported, and may provide some insight into the pitfalls of complex reconstructive surgery in the adult. For example, in a series of adults who underwent adult hypospadias repair, redo operations had a worse outcome than primary cases (6).

Adult stricture patients with previous hypospadias repair differ from a usual population of stricture patients. First, they sometimes had no voiding complaints even when their strictures were severe. Second, they had complicating problems seldom seen in other stricture patients, including complete renal failure and urethral fistula. Third, they have a poor quality of tissue which requires more complex repairs such as first-stage Johanson operations, with buccal grafts placed in the first stage, followed by second-stage closure later. The associated complications do represent a factor influencing the surgical strategy. However, major determinants were the stricture length, availability of healthy tissue, as well as surgeon's own preference. In patients with long stricture and prominent scarring, we suggested staged repairs if formal reconstruction was planned. Last, they often have such long and hopeless abnormal anterior urethras, that is, by both patient and surgeon, it is determined best to treat them expediently with simple perineal urethrostomy instead of formal repair. In this way, reliable egress of urine can be virtually guaranteed after a short 1–2 hour operation, an option chosen by 5/9 (56 %) of our cases. Our experience here exemplified that heroic measures were not always justified to treat the severest urethral strictures, and that perineal urethrostomy can be a gratifying option.

Repair of posthypospadias strictures in children has been widely discussed by pediatric urologists. Modern series now favored single-stage, two-stage buccal mucosal graft repair, or urethroplasties utilizing tunica vaginalis (10, 11). Urethral stricture is a known complication following hypospadias repair (7–9), but the true incidence is unknown. Some childhood hypospadias series do not follow the patients long enough to report any strictures, and when series do report strictures, they usually report them as acute events that occur while the patient is still in childhood, not later as adults. A series by Duel et al., for example, showed a stricture incidence as high as 6.5 % (38 of 582) after pediatric hypospadias surgeries (3). They demonstrated that strictures occurred after a (mean) interval of 27 months. 79 % of these pediatric urethral strictures ultimately required open urethroplasty for correction, and they had a 78 % overall successful rate (12).

I agree, and tend to offer two-stage buccal mucosal repairs such as described by Johanson in adults. We also acknowledge

that some of these patients have such extensive disease, and little interest in a two-stage operation to fix the problem, and thus are most appropriately treated with a perineal urethrostomy. Perineal urethrostomy was offered as a second choice in addition to formal urethral repair. Comorbidity and previous failed urethroplasty were the major factors influencing patients who accepted perineal urethrostomy.

Conclusion

Honestly I believe that this is nothing but an assumption and time is the vital deadline here and it will take more years of debate and research before an original fruitful final conclusion is reached, of course. I am quite certain about the fact indeed and it is the reality without any doubt.

Urethral stricture may occur decades after initial hypospadias surgery. It can be the most severe form of anterior urethral stricture, and may eventually require salvage treatment such as a perineal urethrostomy. Patients undergoing hypospadias surgery should receive lifelong follow-up protocol to detect latent urethral strictures. Patient often have few voiding complaints and can present with severe complications. Two-stage urethroplasties with buccal mucosal grafts can achieve good result when necessary. The stricture can be very extensive and may require salvage treatment such as a perineal urethrostomy. Urethral stricture can occur decades after initial hypospadias surgery. We suggest that patients undergoing hypospadias surgeries should receive lifetime follow-up to detect latent urethral strictures, and that research reports discussing stricture after hypospadias repair include very-long-term follow-up data to determine the exact incidence of this problem.

So, it is very clear that though the study was conducted in a very small sample of population at the Internal Medicine Indoor Department, Dhaka Medical College Hospital, Bangladesh, it may be unable to depict the more realistic picture in this connection as a whole, that is – in fact the actual situation may be more severe than it is depicted here. Moreover not any satisfactory number as well as level of study in this relation is available in our developing country until now. So the most important pearl is that indeed study in large scale should badly require just now having a more realistic and a more accurate image of this burning as well as alarming problem just now.

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