Observational retrospective study on acquired megalourethra after primary proximal hypospadias repair and its recurrence after tapering

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Abstract
Introduction: Acquired megalourethra (AMU) after repair of proximal hypospadias can be a serious complication. An observational retrospective study of its incidence among different types of repair was performed.

Materials and methods: Clinical charts of patients operated on for proximal hypospadias were reviewed. Inclusion criteria: all primary hypospadias operated in 1991–2004, with the meatus positioned in proximal penile, scrotal or perineal position.

Results: Of 770 hypospadias cases treated, 130 (16%) were proximal. Seventy-two patients (55%) were treated using preputial flaps: 36 with a tubularized preputial island flap (TIF) and 36 an onlay island flap (OIF). Fifty-eight patients (45%) underwent staged repairs: Belt-Fuquà (BF) in 18 and Bracka procedure in 40 cases. After a mean follow up of 16 years (range 6–19) the overall incidence of complications for each technique was: TIF 36%; OIF 33%; BF 25%; two-stage Bracka 7.5%. The most common complication encountered was neo-urethral fistula. AMU occurred in only 5 cases, none with associated distal urethral stenosis, all in the TIF and OIF groups, and all successfully treated by reduction re-do urethroplasty.

Conclusion: A very small number of the patients operated using preputial island flaps techniques developed AMU. None of the staged repairs developed AMU, and this is the preferred choice in proximal hypospadias when the urethral plate requires division and/or substitution. All cases of AMU resolved after urethral tapering.

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Introduction

Multiple hypospadias correction techniques have been developed over the years. Some techniques performed many years ago seemed to provide good initial results but unexpected complications occurred with longer follow up. In the 1980s, Duckett and Standoli [1,2] each proposed a single-stage procedure to correct proximal hypospadias based on transferred flaps from the preputial hood, but a significant number of patients developed dilation of the neo-urethra without apparent associated stenosis of the distal urethra. Bracka proposed a two-stage repair using a free graft in the first stage to overcome the problem of late dilation of the neo-urethra [3]. The purpose of the present study was to compare the outcomes of single-stage procedures using preputial flap repairs with the staged techniques utilizing free grafts.

Materials and methods

After approval of the local Institutional Review Board, a retrospective observational analysis of consecutive patients treated for primary proximal hypospadias by a single surgeon (GM) from January 1991 to December 2004 was performed. Surgical techniques and complications were recorded for each case, focussing on the development of acquired megalourethra (AMU) and urethrocutaneous fistulas.

The study was reported according to the STROBE statement (Strengthening the Reporting of Observational Studies in Epidemiology) [4]. Only descriptive statistical analysis was used as this was an investigative retrospective observational study.

Results

Of 770 hypospadias cases treated, 130 (16%) were proximal. Seventy-two patients (55%) were operated on using preputial flaps: 36 tubularized island flap (TIF) and 36 onlay island flap (OIF). Fifty-eight patients (45%) underwent staged repairs: Belt-Fuqua (BF) in 18 cases and Bracka procedure in 40 cases (Table 1).

Associated correction of penile curvature by dorsal corporoplasty was performed in 20 patients: 9 of the TIF group (6 by tunica albuginea plication, 3 by Nesbit repair) and 11 of the Bracka group (5 by tunica albuginea plication and 6 by Nesbit); none of the patients of OIF or BF groups required a corporoplasty.

After a mean follow up of 16 years (range 6–19) the overall incidence of complications observed for each technique was: TIF 36% (13/36); OIF 33% (12/36); BF 25% (4/18); two-stage Bracka 7.5% (3/40) (Table 2). Mean follow up was different for the different technique groups being 16 years for the preputial flaps and 12 years for the two-stage repairs.

At the time of this survey the mean age of the patients treated for proximal hypospadias was 15 years (range 7–27), equally distributed among preputial flap and two-stage groups. Sixty-six of them were already post-pubertal: 55 in the preputial flap group and 11 in the two-stage group.

The most common complication encountered in each group was a neo-urethral fistula that occurred in 10, 10, 4 and 3 cases of TIF, OIF, BF and Bracka groups, respectively. No case of recurrence of curvature was observed in the entire series.

AMU occurred in only 5 cases and all of these arose in the island flap groups (3 in TIF, 2 in OIF). There was no evidence of an associated distal urethral stenosis in any case. The clinical presentation of AMU (Figs. 1 and 2) included ventral penile ballooning and post-voiding dribbling, and all were diagnosed in the pre-pubertal age group, at a mean follow up of 7 years (range 3–8) following surgery.

Repair of AMU was successfully performed by urethral tapering in all cases: a portion of the redundant ventral urethra was de-epithelialized and plicated over the tapered urethra in order to provide some mechanical reinforcement and protection against fistula formation (Fig. 3). After 9.33 years of subsequent follow up (range 5–15), no recurrence of AMU was observed.

Discussion

AMU secondary to preputial flap urethroplasties for proximal hypospadias is a serious complication, occurring in 4–12% of cases [5–8], and the 7% in this series reinforces that experience even with longer follow up. As in our series, the majority of AMU cases reported in the literature are not associated with meatal stenosis, suggesting that this complication is likely due to an inherent lack of support tissue of the neo-urethra [7,8]. It could be hypothesized that with a ventral single-stage augmentation technique (OIF, TIF) the flap’s lack of attachment to the surrounding tissues makes the neo-urethra more elastic and thus prone to progressive dilation. The increased diameter of this urethral section may lead to turbulent flow and an increase

| Table 1 | Distribution of cases by type of procedure. |
|---|---|---|
| 1991–2004 | 770 hypospadias | 130 (16%) proximal |
| Single-stage | Two-stage |
| 72 cases | 58 cases |
| 36 (50%) TIF | 18 (31%) Two-stage Belt-Fuqua |
| 36 (50%) OIF | 40 (69%) Two-stage Bracka |

| Table 2 | Incidence and distribution of type of complication by each surgical technique. |
|---|---|---|
| Complication | n (%) | Fistula | AMU |
| TIF | 13 (36) | 10 | 3 |
| OIF | 12 (33) | 10 | 2 |
| 2-stage Belt-Fuqua | 4 (25) | 4 | — |
| 2-stage Bracka | 3 (7.5) | 3 | — |
in the tension of the urethral wall (as explained by Laplace’s law) that further increases dilation with time.

In our series, AMU was successfully repaired by reductive urethroplasty as described by Radojicic et al. [9] and no recurrence was observed after a mean follow up of 9.33 years. The absence of recurrence can be explained by the hypothesis that, similarly to staged techniques, the reconstructed neo-urethral tissue has had enough time to develop a more stable binding with the corporeal bodies and this binding is not disturbed by the revision urethroplasty. Another factor that may contribute to avoiding recurrent urethral dilation is the presence of a midline ventral suture that (as for staged procedures) represents a sort of mechanical support aided by a second layer wrap of de-epithelialized tissue.

Figure 1 Clinical features of AMU with presence of penile mass during and/or after voiding (a, b) and its aspect on voiding cystogram (c).

Figure 2 Intraoperative demonstration of absence of meatal stenosis (a, b) and urethral dilatation (c, d).

Figure 3 Sequence of surgical procedure for AMU: sagittal penile incision to expose diverticula (a) that was then completely opened through ventral incision (b, c); de-epithelialization of the dilated urethral wall (d, yellow dashed area) and urethral tapering using the de-epithelialized area as a mechanical reinforcement for the neo-urethra and protective flap for fistula (e). Final result (f). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)
In some of the cases reported in the literature there is a mild misalignment of the neo-urethra detected on urethrography that, however, seems not to cause impairment of urinary stream [6]. These findings were not observed in our series.

In the present series, the one-tailed Fisher’s exact test was statistically significant for the incidence of AMU in flap techniques (p = 0.0488) while the bidirectional test was less significant (0.0649). Even if it could be related to the dimension of the sample population it shows a trend that, in our opinion, cannot be ignored (Table 3).

There was no statistically significant difference in the incidence of urethrocutaneous fistula among the two groups (p = 0.183) although there was a tendency to develop fewer fistulas in the Bracka technique group (7.5% compared to 36% and 33% in the flap groups) (Table 3). No other neo-urethral coverage than peri-urethral tissue was used in both groups. Probably the different results between the one-stage and the two-stage groups may be related to a more healthy tissue provided by staged techniques that allows the surgeon to make a more reliable coverage.

Difference in follow up duration among groups in this series (mean 16 years for the preputial flap group and 12 years for the two-stage group) is related to the evolution in the surgical strategy from an initial extensive application of preputial flap techniques to an almost exclusive use of Bracka two-stage technique in the last period of this series. This may create a bias in the different incidence of complications due to the learning curve factor, but we consider that it is not enough to explain the complete absence of AMU in the two-stage group.

The present paper underlines the limitations of an observational retrospective study. Bias may be related to the authors’ recent preference to manage most of the cases of proximal hypospadias with staged repairs. This preference affects principally the last 5 years of the study period and was mainly related both to better cosmetic (principally in terms of a slit meatus) and functional outcomes together with a decreased rate of fistula formation (up to 36% with flaps versus 7.5% with grafts). While for AMU and fistulas there are data that can be analysed, the cosmetic appearance is a totally subjective factor for which there are no reliable data and for this reason it can’t be compared by statistical analysis.

This paper is focused on the development of AMU as a matter of record, and it would not be appropriate to reduce this discussion to one of argument between staged versus single-stage techniques as a matter of principle. Even if Mureau et al. found no significant correlation between psychosexual adjustment and the number of operations in their study [10] it is advisable to reduce the number of surgical procedures to a minimum. It can be argued that the staged techniques are a “scheduled complication”. In the present study the average number of surgeries per technique, including complications, was higher in the staged than in the flap techniques (TIF 1.36; OIF 1.33; BF 2.22; Bracka 2.08). However, in our experience, there is a marked difference between a straightforward second stage procedure and the correction of an urethral fistula, making the latter a more complicated surgery to be avoided as often as possible.

Conclusion

In our series postoperative AMU has occurred only when preputial flap techniques were utilized for severe proximal hypospadias repairs. When compared with the two-stage group statistical analysis showed a significant difference. In addition the superior cosmetic results and the lower incidence of fistula formation (even if no statistical data are available), support the authors’ preference for staged repairs using dorsal free grafts for the management of proximal hypospadias when division of the urethral plate is necessary.

Conflict of interest

None.

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References