Journal of Pediatric Urology (2017) 13, 35.e1–35.e7

Two-stage hypospadias repair with a free graft for severe primary and revision hypospadias: A single surgeon's experience with long-term follow-up

K.L.M. Pfistermüller, S. Manoharan, D. Desai, P.M. Cuckow

Department of Paediatric Urology, Great Ormond Street Hospital, Great Ormond Street, London, WC1N 3JN, United Kingdom

Correspondence to: K.L.M. Pfistermüller, Department of Paediatric Urology, Great Ormond Street Hospital, Great Ormond Street, London, WC1N 3JN, United Kingdom, Tel.: +44 7889 650080

klmpfist@aol.com
(K.L.M. Pfistermüller)

Keywords
Hypospadias; Two-stage repair; Long-term follow-up; Outcomes

Received 13 January 2016
Accepted 17 August 2016
Available online 29 September 2016

Summary

Introduction
Repair of severe primary and revision hypospadias is a demanding procedure. Debate continues as to whether a two-stage approach or single-stage technique is superior. The two-stage procedure with a free graft involves penile straightening followed by application of a graft for the neourethral plate at stage one; with tubularization at stage two after graft maturation.

Objective
To report the outcomes of a single surgeon's experience with the two-stage repair using a free graft for both severe primary and revision hypospadias with long-term follow-up.

Materials and methods
Between July 1998 and January 2010, 301 boys underwent a two-stage reconstruction. The surgical technique is described in the manuscript.

Primary repairs ($n = 208$): indications for a two-stage approach with a free graft included meatal position, presence of corporal chordee, and poor glans development. Median follow-up from completion of the second stage was 75 months.

Revision repairs ($n = 93$): indications included urethral fistula, excessive scarring/meatal stenosis, balanitis xerotica obliterans (BXO), and residual or untreated chordee. Median follow-up from completion of the second stage was 85 months.

Results
For the primary repairs ($n = 208$), the graft took well in all but one case. Second-stage complications included fistula (7), meatal stenosis (3), partial glans dehiscence (3), and all were re-operated (13).

For the revision repairs ($n = 93$), the graft took well in all but four cases. Second-stage complications included fistula (5), meatal stenosis (3), breakdown (1) and reoperation (8).

Discussion
In a systematic review of 20 years of publications on the repair of primary severe hypospadias, the two-stage procedure with a free graft demonstrated an overall complication rate of 22%; this was a distinct overall benefit when compared with the single-stage procedures in terms of lower complication rates (Castagnetti and El-Ghoneimi, 2010). Our results for the severe primary repairs revealed significantly lower complication rates than those in the literature, with an overall re-operation rate of 6.3%, a fistula rate of 3.4%, and meatal stenosis and partial glans dehiscence at 1.4% each.

Several papers have documented outcomes following the single-stage tubularized incised plate urethroplasty for re-operative hypospadias, giving overall complication rates ranging from 15.4 to 30%. Our data show a re-operative rate of 8.6%, a fistula rate of 5.3%, breakdown in 1.1%, and meatal stenosis in 3.2%.

Conclusion
The two-stage repair with a free graft for correction of both severe primary and failed primary hypospadias is a safe, viable, and durable procedure offering low morbidity and excellent cosmetic results. The authors advocate the two-stage repair with a free graft as the technique of choice for treatment of both of these challenging groups of the deformity.

Summary Table Graft type for primary and revision repairs.

Graft type	Primary repairs ($n = 208$)	Revision repairs ($n = 93$)
Inner prepuce	199	15
Posterior auricular Wolfe graft	4	60
Buccal mucosal graft	1	9
Composite graft	4	6
Shaft skin	0	3

http://dx.doi.org/10.1016/j.jpurol.2016.08.014

Introduction

Repair of severe primary and revision hypospadias is a demanding procedure. There is ongoing debate as to whether a two-stage approach or single-stage technique is superior. Castagnetti et al. recently published a systematic review of 20 years of publications on severe primary hypospadias management; lower complication rates were shown with a staged approach [1]. Steven et al. surveyed current practice in paediatric hypospadias surgery, and showed that nearly half of all respondents opted for a staged approach with a free graft for correction of proximal hypospadias [2].

Turner-Warwick and Cloutier should be acknowledged for their initial descriptions of staged urethral reconstruction for both primary and salvage procedures [3,4]. More recently, Bracka re-popularised this technique [5]. The procedure involves penile straightening followed by application of a graft for the neourethral plate at stage one; with tubularization at stage two after graft maturation. When applied to the revision repairs, the technique allows for complete excision of scar tissue from previous procedures prior to laying a fresh graft of either harvested skin or buccal mucosa.

The present study reports the outcomes of a single surgeon's experience with the two-stage repair with a free graft for both severe primary and revision hypospadias with long-term follow-up.

Materials and methods

Between July 1998 and January 2010, 301 boys underwent a two-stage hypospadias reconstruction using a free graft: 208 were primary repairs and 93 revisions.

Primary repairs

For the primary repairs, the pre-operative meatal position is detailed in Table 1, although this was not the exclusive measure of severity or the prime indication for a two-stage approach. Other associated factors included the presence of corporal chordee (as assessed during surgery) and poor glans development. A proximal meatus was defined as sited anywhere from the proximal shaft to the perineum. No pre-operative hormonal stimulation was given to any patient. The median age at first operation was 16 months (range: 10—204). Median follow-up from completion of the second stage was 75 months (range: 12—137).

Table 1 Pre-operative meatal position.

Pre-operative meatal position	Primary repairs $n = 208$	Secondary repairs $n = 93$
Glans	4	11
Coronal	7	5
Subcoronal	0	11
Distal shaft	19	25
Mid shaft	51	15
Proximal shaft	29	10
Penoscrotal	87	16
Perineal	11	0

Revision repairs

These came from a referral practice, and the indications included urethral fistula, excessive scarring/meatal stenosis, balanitis xerotica obliterans (BXO), residual or untreated chordee, and previous breakdown. Median age at first operation was 86 months (range: 7—280). Median follow-up from completion of the second stage was 85 months (range: 12—137).

Surgical technique

At the beginning of the first stage, the penis is degloved circumferentially to the level of Buck's fascia. Ventrally, the urethral plate is divided and lifted together with the spongiosum and the proximal urethra from the ventral aspect of the penile corpora. This continues proximally to the bifurcation of the corpora and releases them completely to optimise their length. At this point, an artificial erection is performed to assess residual penile chordee, using a proximal tourniquet and normal saline (Fig. 1). If present, chordee is corrected by lifting the neurovascular bundle off the corpora and performing dorso-lateral plications of the tunica albuginea, as described by Duckett [6] (Figs. 2 and 3).

Following chordee correction, to create a deep glans groove, the glans is split in the midline. Further dissection of the glans, off the dome of the corpora, is performed to allow the glans to open out flat [3]. This allows a rectangular graft shape that will provide easy tubularisation at

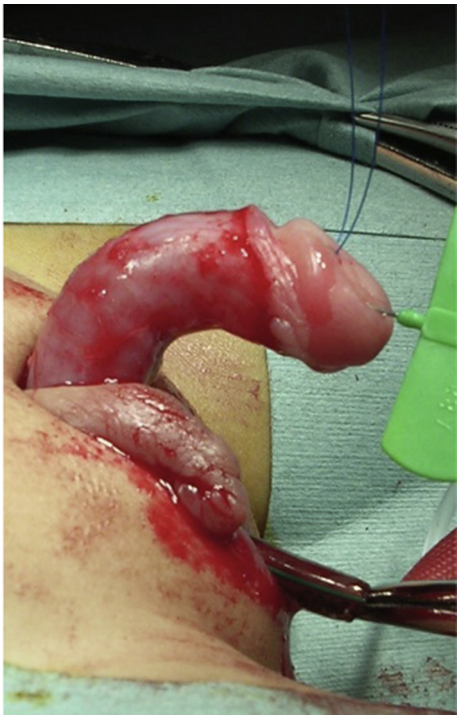

Figure 1 Artificial erection to assess residual chordee following penile degloving and urethral plate division.

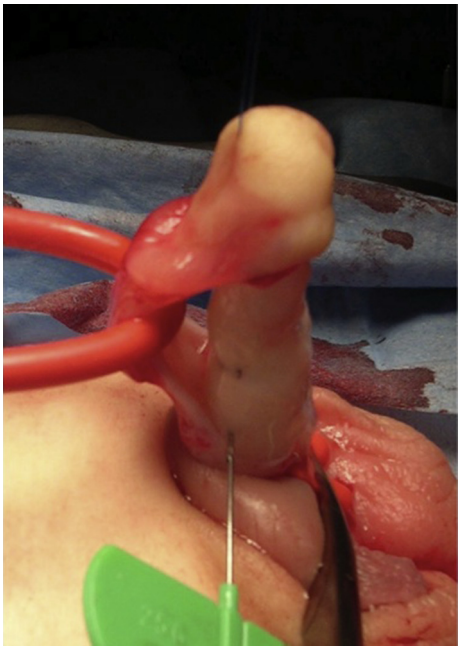

Figure 2 The neurovascular bundle has been dissected off the corpora.

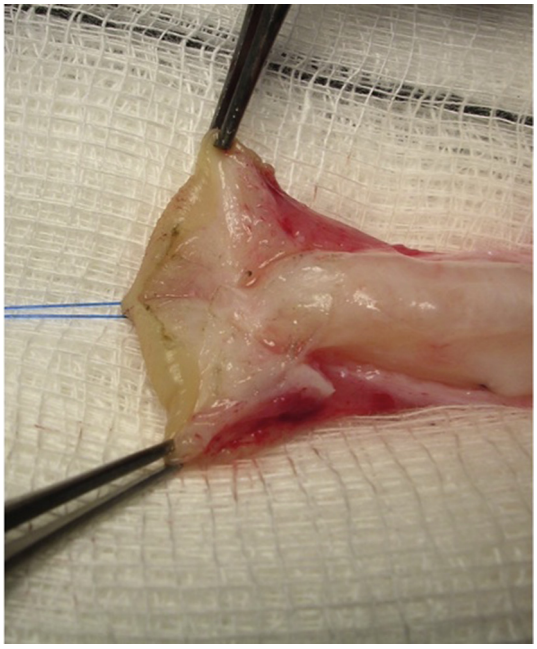

Figure 4 Dissection of the glans off the dome of the corpora allowing the glans to open out flat.

the second stage (Figs. 4 and 5). When applied to revision procedures, the failed neourethra is either partially or completely excised back to healthy bleeding tissue. In addition, in the revision repairs with meatal stenosis, a temporising proximalisation of the urethral opening may have been performed prior to planned repeat repair.

The graft is harvested, usually inner preputial skin, to leave sufficient skin for coverage of the shaft. When the inner preputial skin is deficient (due to previous repair, circumcision or balanitis xerotica abliterans), an extragenital graft is harvested (Table 3). Anastomosis of the shaft skin to the mucosal cuff of the glans is then performed to achieve dorsal and lateral coverage of the shaft.

The remaining raw ventral area is held open with sutures, and the cleaned graft applied. To hold in place, sutures are inserted to the periphery of the graft joining it proximally to the glans, preputial skin and spongiosum.

Figure 3 Artificial erection following chordee correction.

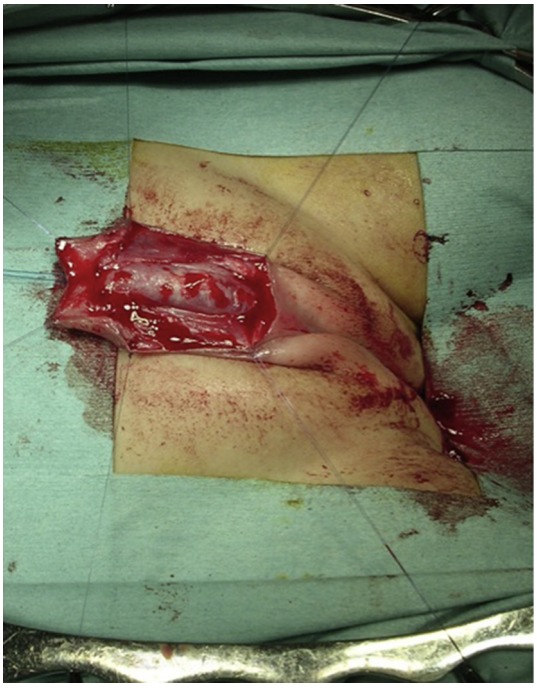

Figure 5 The rectangular graft bed.

Table 2 Complications after second stage for primary repairs.

Complication	Patient number (n = 208)	Treatment
Fistula	7	Fistula repair
Meatal stenosis	3	Meatotomy
Partial glans dehiscence	3	Glanuloplasty
Penoscrotal tethering	1	Observation
Deviated stream	2	Observation
Post micturition dribble	2	Observation
Mild residual chordee	3	Observation

Table 3 Number of previously failed repairs performed elsewhere prior to secondary repair by senior author.

Number of previously failed repairs	Number of patients (n = 93)
1–4	63
5–9	25
10–14	3
15–20	2

Quilting sutures are used to secure the graft to the shaft, and small perforations are made to release any haematoma (Fig. 6). A roll of paraffin gauze is placed on to the graft, and the shaft skin sutured around it to give compression of the graft onto the penis (Fig. 7). A proximal dripping stent is placed in the bladder and secured with a suture. The penis is then dressed with a foam pressure dressing (Cavi-care, Smith and Nephew) (Fig. 8). The child is admitted for one night, and Augmentin and Oxybutynin are given for one week postoperatively.

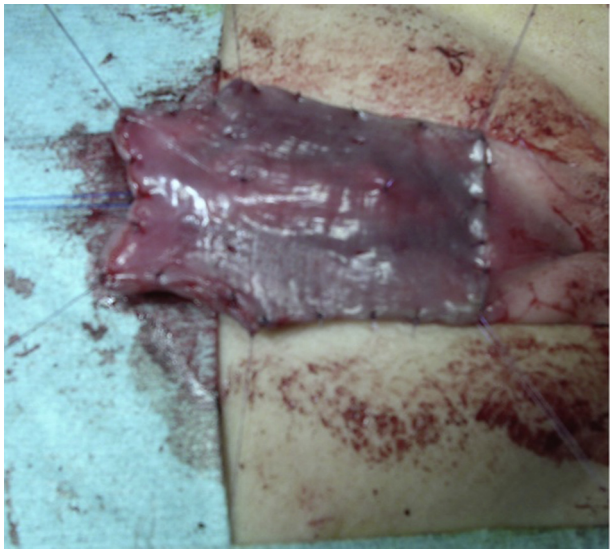

Figure 6 The free graft in place and held with peripheral sutures to secure it to the glans, preputial skin and spongiosum proximally. Quilting sutures are used to secure to the shaft and small perforations made to release any haematoma.

Figure 7 A roll of paraffin gauze is placed on to the graft and the shaft skin sutures around it to give compression.

After 1 week, the child returns for removal of the dressing under general anaesthetic, when the holding sutures are removed and the graft is assessed. Topical Chloramphenicol ointment is applied to the graft, and this

Figure 8 Dripping stent and compression dressing.

is continued twice daily for 1 week. The child is brought for outpatient review at 3 and 6 months postoperatively, and scheduled for completion surgery ensuring at least 6 months have elapsed between stages.

At the second stage, tubularization of the neourethra, an incision is made on either side of the graft where it joins the shaft skin and this is tubularized around an 8-Ch feeding tube (Figs. 9 and 10). A second waterproofing dartos layer is interposed between this and the skin closure (i.e. a three-layered closure technique). The same dressing and after-care medication is applied as for the first stage, with removal on the ward after 1 week. The child is then reviewed at 3 and 12 months, with ongoing annual review.

Results

Primary repairs

Inner preputial skin was the graft of choice — it was used in 199 of the 208 cases. In cases where insufficient inner preputial skin was available, a posterior auricular Wolfe graft (PAWG) was used in four cases, buccal mucosa in one case, and a composite graft of inner preputial skin and PAWG in four.

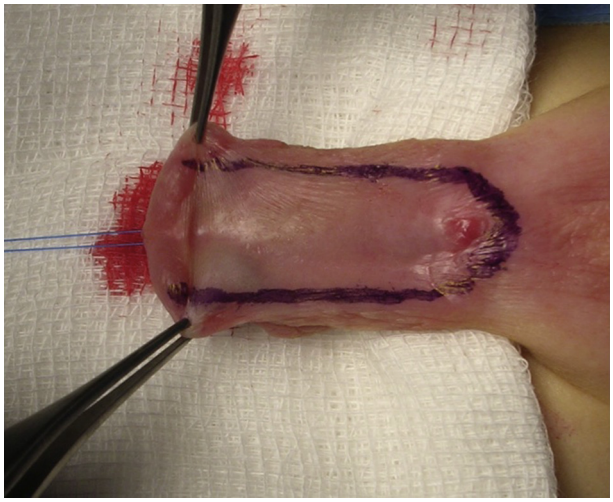

Figure 9 Markings for incision of the graft.

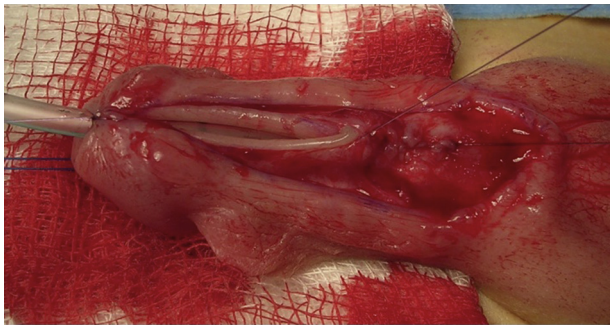

Figure 10 Tubularization of the neourethra over an 8-Ch feeding tube.

The graft took well in all but one case, which required revision. Two cases suffered mild focal scarring, which was excised at the second stage. Four developed a haematoma beneath the graft, which was evacuated and redressed for another week. The subsequent result for these six cases that were treated conservatively was excellent.

Cosmetic and functional outcomes after the second stage were excellent in 187, good in eight, and 13 required re-operation. Outcome was assessed by both the surgeon's and parents' assessment of function and cosmetic appearance. Complications after the second stage were: seven fistulae (3.4%), three meatal stenosis (1.4%), and three partial glans dehiscence. All of these were treated with repeat surgery, giving a re-operation rate of 6.3%.

In the eight cases where outcome was described as good, one had mild penoscrotal tethering, three had mild residual chordee, two had a deviated urinary stream, and two suffered with post micturition dribble. All of these were treated conservatively with observation (see Table 2).

Revision repairs

None of the previous failed repairs were originally performed by the senior author. Table 3 demonstrates the number of previous repair attempts prior to definitive repair. A posterior auricular Wolfe graft (PAWG) was the graft of choice in 60 cases. Where sufficient inner preputial skin was available, this was used in preference and was the graft material in 15 cases in this series. Shaft skin was used in three, and a composite graft of shaft or inner preputial skin with a PAWG in six. A buccal mucosal graft (BMG) was used in nine. The grafts took well in 89 of the 93 cases. In three cases, where a PAWG was used, keloid developed in the graft and two of these cases required revision. In one case, the BMG shrunk and re-grafting was performed with a PAWG. One child developed an implantation dermoid (excised at stage two) and three children developed a haematoma under the graft (managed as for the primary repairs). Two children had dehiscence of the PAWG donor site, which was managed conservatively; one developed a keloid scar at the donor site.

Cosmetic and functional outcomes after the second stage, as judged by the surgeon, parents and patients (where old enough), were excellent in 84 cases. Eight children (8.6%) required re-operative intervention: five for a fistula (5.3%) (one was associated with meatal stenosis);

Table 4 Complications after second stage for secondary repairs.

Complication	Patient number (n = 93)	Treatment
Fistula	5	Fistula repair
Breakdown	1	Repeat staged procedure
Meatal stenosis	3	Meatotomy
UTI	2	Antibiotics
PAWG site dehiscence	2	Conservative

PAWG, posterior auricular Wolfe graft.

one had breakdown (1.1%); and three had meatal stenosis (3.2%). Two patients developed a UTI following the second stage, and one of these was diagnosed with underlying reflux (see Table 4).

Discussion

Repair of severe primary hypospadias remains a significant surgical task for which no one technique is universally applied. Results of a worldwide survey evaluating trends in hypospadias surgery revealed that the two-stage technique using a free graft was preferred by 43.3—76.6% of respondents for the repair of proximal hypospadias [7]. A similar trend favouring a two-stage free graft repair for proximal hypospadias was demonstrated in the results of another specialist survey showing that 49% would perform a two-stage repair; 15% opted for the single-stage tubularized incised plate urethroplasty [2]. In a systematic review of 20 years of publications on the repair of primary severe hypospadias, the two-stage free graft procedure demonstrated an overall complication rate of 22%, with a 10% fistula/dehiscence rate, and a 6% stricture/stenosis rate. This revealed a distinct overall benefit when compared to the single-stage procedures in terms of lower complication rates, but the authors commented on committing the child to a second surgical procedure. Of note, this systematic review also commented that the only long-term outcome papers were the two-stage free graft reconstructions (due to the popularity of this technique in the 1980s), although no comment was made on the average follow-up duration for the single stage repairs [1]. Attention should be drawn to a recent publication looking at long-term follow-up after repair of all degrees of primary hypospadias. This revealed that 47.4% of the 24.1% cases requiring re-operative intervention presented within the first 12 months postoperatively [8]. A comparison of the tubularized incised plate urethroplasty with the onlay island flap urethroplasty for penoscrotal hypospadias revealed overall complication rates of 60% and 45%, with rates of fistula at 42.9% vs 20%, break down at 8.6% vs 5%, and residual chordee at 5.7% vs 12.5%, respectively. There was a mean follow-up of 30 months for the tubularized incised plate urethroplasty and 38.8 months for the onlay island flap [9]. Outcomes for the Macedo 3-in-1 technique for repair of complex primary hypospadias (reconstruction of the urethral plate with buccal mucosa followed by an onlay transvers preputial flap and waterproofing tunica vaginalis flap as a single-stage procedure) were a mean follow-up of 55.2 months, an overall complication rate of 37%, and a re-operation rate of 31.5%. Rates of meatal stenosis and urethral diverticula were given as 11.4% with fistulae at 14.3% and residual chordee at 5.7% [10]. Results for the severe primary repairs from the present patient cohort, with a mean follow-up of 72.3 months, revealed significantly lower complication rates than those demonstrated in the Castagnetti review paper for single and two-stage repairs, as well as the Braga and Macedo papers [1,9,10], with an overall re-operation rate of 6.3%, a fistula rate of 3.4%, and meatal stenosis and partial glans dehiscence at 1.4% each.

Management of failed primary/revision repair or the so called 'hypospadias cripples' poses even more of a reconstructive challenge due to the already scarred tissues and limited reconstructive options. Several papers from the 2000s have documented outcomes following the single-stage tubularized incised plate urethroplasty for re-operative hypospadias, giving overall complication rates ranging from 15.4% to 30% and fistula rates of 7.7—20% [11—14]. Application of the Mathieu repair to salvage hypospadias has given poorer outcomes than the tubularized incised plate urethroplasty, with overall complication rates at 22.5—30.9% and fistula rates of 15—25.5% [15,16]. A recent study by Gill et al. reviewed 100 cases of re-operative hypospadias repair using a staged approach [17]. This study revealed a 13% re-operation rate, 9% fistula rate, 6% urethral stricture rate, and 1% glans dehiscence. The paper concluded that the two-stage free graft approach should be the first-line treatment for hypospadias cripples due to superior results, versatility and reproducibility. The present data have shown a re-operative rate of 8.6%, a fistula rate of 5.3%, breakdown in 1.1% and meatal stenosis in 3.2%. This gives results similar to the present outcomes for primary severe hypospadias, and the authors are in agreement with Gill et al. in recommending a two-stage free graft approach as the definitive treatment for correction of failed primary hypospadias repair.

Limitations

The limitation of this study was that it was a retrospective review. However, the data were strong, as it was from a single surgeon and performed using the same technique with all patients followed up according to a set protocol. Whilst no subjective scoring system was used, or available during the early part of this practice, an assessment of all the stated outcome measures was performed by the primary surgeon, his junior doctors, experienced specialist nurses, the parents and, most importantly, the patients themselves, at each clinic follow-up visit.

Conclusion

The two-stage free graft repair for correction of both severe primary and failed primary hypospadias is a safe, viable and durable procedure offering low morbidity and excellent cosmetic results. It is advocated that the two-stage free graft repair should be used as the technique of choice for treatment of both of these challenging groups of the deformity.

Ethical approval

The project was registered and approved by the Research and Development department at Great Ormond Street Hospital.

Funding sources

None.

Conflict of interest statement

None.

References

[1] Castagnetti M, El-Ghoneimi A. Surgical management of primary severe hypospadias in children: systematic 20-year review. J Urol 2010;184:1469—75.

[2] Steven L, Cherian A, Yankovic F, Mathur A, Kulkarni M, Cuckow P. Current practice in paediatric hypospadias surgery; a specialist survey. J Pediatr Urol 2013;9(6 Pt B): 1107.

[3] Turner-Warwick R. Observations upon techniques for reconstruction of the urethral meatus, the hypospadiac glans deformity and the penile urethra. Urol Clin North Am 1979;6: 643—55.

[4] Cloutier AM. A method of hypospadias repair. Plast Reconstr Surg 1962;30:368—73.

[5] Bracka A. Hypospadias repair: the two-stage alternative. Br J Urol 1995;76(s3):31—41.

[6] Baskin LS, Duckett JW. Dorsal tunica albuginea plication for hypospadias curvature. J Urol 1994;151:1668—71.

[7] Springer A, Krois W, Horcher E. Trends in hypospadias surgery: results of a worldwide survey. Eur Urol 2011;60:1184—9.

[8] Spinoit A, Poelaert F, Groen L, Laecke E, Hoebeke P. Hypospadias repair at a tertiary care centre: long-term follow-up is mandatory to determine the real complication rate. J Urol 2013;189:2276—81.

[9] Braga LHP, Pippi Salle JL, Lorenzo AJ, Skeldon S, Dave S, Farhat WA, et al. Comparative analysis of tubularized incised plate versus onlay island flap urethroplasty for penoscrotal hypospadias. J Urol 2007;178:1451—7.

[10] Macedo Jr A, Liguori R, Ottoni SL, Garrone G, Damazio E, Mattos RM, et al. Long-term results with a one-stage complex primary hypospadias repair strategy (the three-in-one technique). J Pediatr Urol 2011;7:299—304.

[11] Snodgrass W, Lorenzo A. Tubularized incised-plate urethroplasty for hypospadias reoperation. Br J Urol Int 2002;89: 98—100.

[12] Shanberg A, Sanderson K, Duel B. Re-operative hypospadias repair using the Snodgrass incised plate urethroplasty. Br J Urol Int 2001;87:544—7.

[13] Nguyen M, Snodgrass W. Tubularized incised plate hypospadias reoperation. J Urol 2004;171:2404—6.

[14] Ziada A, Morsi H, Aref A, ElSaied W. Tubularized incised plate (TIP) in previously operated (redo) hypospadias. J Pediatr Urol 2006;2:409—14.

[15] Bar-Yosef Y, Binyamini J, Matzkin H, Ben-Chaim J. Salvage Mathieu urethroplasty: reuse of local tissue in failed hypospadias repair. Urology 2005;65:1212—5.

[16] Emir L, Erol D. Mathieu urethroplasty as a salvage procedure: 20-year experience. J Urol 2003;169:2325—7.

[17] Gill N, Hameed A. Management of hypospadias cripples with two-stage Bracka's technique. J Plast Reconstr Aesthetic Surg 2001;64:91—6.