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# One-stage combined delayed bladder closure with Kelly radical soft-tissue mobilization in bladder exstrophy: preliminary results



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#### Summary

#### Background

The radical soft-tissue mobilization (RSTM, or Kelly repair) is an anatomical reconstruction of bladder exstrophy generally performed as a second part of a two-step strategy, following successful neonatal bladder closure.

#### Objective

The objective of this study is to determine the feasibility of a combined procedure of delayed bladder closure and RSTM in one stage without pelvic osteotomy, in both primary and failed initial closure.

## Design, setting, and participants

From 11/2015 to 01/2018, 27 bladder exstrophy patients underwent combined bladder closure with RSTM by the same surgical team at four cooperating tertiary referral centers for bladder exstrophy, including 20 primary repairs (delayed bladder closure, median age 3.0m [0.5-37m]) and seven secondary repairs after failed attempt at neonatal closure, median age 10m [8-33m].

#### Intervention

RSTM included full mobilization of the bladder plate, urogenital diaphragm, and corpora cavernosa from the medial pelvic walls, followed by anatomical reconstruction with antireflux procedure, bladder closure, urethrocervicoplasty, muscle sphincter approximation, and penile/clitoral reconstruction.

#### Outcome measurements

The main criteria were bladder dehiscence or prolapse. Secondary outcomes included bladder neck fistula or urethral fistula, urethral stenosis, and parietal hernia. Continence and voiding have not been addressed at this stage.

#### **Results and limitations**

All bladder exstrophy cases were successfully closed without osteotomy, with no case of bladder dehiscence after 12 m [3-30] follow-up.

#### Complications

Urethral fistula or stenosis occurred in eight patients: 4/5 fistulae closed spontaneously in less than 3 months; four urethral stenoses were successfully treated with 1–3 sessions of endoscopic highpressure balloon dilatation or meatoplasty; one patient with persistent bladder neck fistula is currently awaiting repair. Although the follow-up is short, it does allow examination of the main outcome criterion, namely bladder dehiscence, which is usually expected to happen very early after surgery.

#### Conclusion

The Kelly RSTM can be safely combined with delayed bladder closure without osteotomy in both primary and redo cases in classic bladder exstrophy.

Table								
	Primary delayed closure + Kelly	Redo closure + Kelly						
n	20	7						
Males/females	13/7	5/2						
Age (mo.)	3.0 [0.5–37]	10 [8–33]						
Bladder dehiscence	0	0						
Urethral stenosis	1/20	3/7						
Urethral fistula	3/20	2/7						

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# Introduction

Modern techniques in bladder exstrophy reconstruction aim at providing an adequate reservoir and urinary continence. while preserving renal function. Among them, the modern staged repair (MSRE) and the complete primary reconstruction of exstrophy (CPRE) have been the most extensively studied and reported procedures [1,2]. The radical soft-tissue mobilization (RSTM) described by Kelly is an alternative technique aiming at anatomical reconstruction of the bladder neck, urethra, and muscular sphincteric structures in association with a unique mobilization of the corpora cavernosa from the ischiopubic rami to improve penile length in males. It remains a technically challenging procedure, which has been so far uniformly performed in infants and children as the second part of a two-step strategy after successful neonatal bladder closure [3-5]. With the emerging concept of delayed bladder closure [6], the feasibility of a one-stage delayed bladder closure combined with RSTM in children with classic bladder exstrophy was investigated.

# Patients and methods

From November 2015 to January 2018, a single-stage procedure combining bladder closure and Kelly RSTM was performed for classic bladder exstrophy patients, either with untreated bladder template (intentional delayed bladder closure or late referral) or with bladder dehiscence following closure attempt during neonatal period.

# Setting

A total of 27 procedures were performed by a single surgeon (MDL), either alone or as part of a joint team (SF, SS, PGR, GA, YH) in tertiary-care centers of pediatric urology of four cooperating institutions: Nantes Children's University Hospital, France (n = 13), as a referral center for a supraregional healthcare program for bladder exstrophy, Paris Armand Trousseau Children's Hospital (n = 4), Karachi Sindh Institute of Urology and Transplantation, Pakistan (n = 8), and Melbourne Royal Children Hospital (n = 2).

# Technique

Surgical procedure was performed under general anesthesia, supplemented with lumbar epidural continuous analgesia (ropivacain 7 mg/kg/24 h) for 48–72 h postoperatively whenever possible. Surgical steps in male bladder exstrophy are detailed in the accompanying video. The philosophy of the Kelly repair implies two distinct phases: a radical mobilization of the urogenital structures (video 1), including detachment of the anterior pelvic floor from the pelvic bones, followed by a phase of reconstruction (video 2) including antireflux surgery, bladder closure, bladder neck reconstruction, tubularization and ventral transposition of the urethra, penile chordee correction with corporoplasty in males, and penile skin or mons pubis reconstruction. Supplementary video related to this article can be found at https://doi.org/10.1016/j.jpurol.2018.07.013.

# Mobilization

The initial incision was performed around the bladder template and extended along the margins of the urethral plate. It was then continued around the coronal sulcus on the ventral surface and through the scrotum in the midline to the perineum. The area of glabrous skin, constantly seen in males between the scrotum and the penis, was carefully preserved to provide rectangular flaps allowing dorsal penis skin cover. The RSTM was performed using a combined intrapelvic and perineal approach (Fig. 1). Intrapelvic access allowed detachment of the levator ani muscle insertions from the internal obturator fascia, providing access to the ischiorectal fossa and Alcock's canal to facilitate control of the pudendal neurovascular bundles. Perineal dissection allowed exposure of corpora cavernosa, and the use of muscle stimulator facilitated identification of bulbospongiosus, ischiocavernosus, and transversus perinei muscles. The periosteum of the ischiopubic rami was incised and peeled away to allow full mobilization of both corpora which continued anteroposteriorly, until the neurovascular pedicles exiting from the Alcock's canal were identified. All striated muscular structures, part of the urogenital diaphragm, linking the urogenital complex to the anterior pelvic ring, were detached from the bone. Further dissection allowed separation of both corpora from the urethral plate and bulbar spongiosum tissue. In males, if the urethral plate was short and if it was felt that placement of the reconstructed bladder neck within the pelvic cavity would retract the corpora and consequently shorten the penis, the distal urethra was disconnected from the glans to be placed in hypospadiac position which may be on the proximal shaft at the penoscrotal junction or in the perineum.

## Reconstruction

Antireflux procedure was performed either according to the midline Gil-Vernet trigonoplasty technique in case of severely polypoid bladder or preferably with a submucosal cephalotrigonal ureteral reimplant. Excision of full thickness triangles at the level of the bladder neck and tubularization over an 8Fr catheter resulted in a funneled urethrocervicoplasty. Muscular structures mobilized in continuity with the bulbocavernosus muscles were wrapped around the urethra and sutured together dorsally. In cases with only minor pubic diastasis, the pubis was approximated with three mattress sutures. In the majority of cases, who had significant diastasis, the rectus abdominis muscles were detached by subperiosteal incision, allowing both muscles to be sutured together on the midline for a tension-free abdominal closure without the need for osteotomy or pubis approximation. The corpora were anchored to the neo-symphysis using non-absorbable sutures. Ureteral stents and urethral and suprapubic catheters were left in free-drainage for 21 days before bladder cycling. Postoperative immobilization was maintained with a mermaid bandage, a posterior splint (Fig. 2), or a 'frog' plaster cast for 3 weeks.



**Fig. 1** 1A: Schematic view of the inner pelvic floor. Levator ani muscle (a: puborectalis fibers; b: pubococcygeal fibers; c: iliococcygeal fibers; d: sacrospinous ligament); 1B, 1C: Coronal view of the foramen obturatorius (from 1A). Levator ani anterior muscle (e) inserted on the internal obturator (k) fascia. Alcock's canal with pudendal pedicle (l). Ilio-pubic (f) and ischio-pubic (g) rami. Corpus cavernosus (i). Urogenital diaphragm (h). Ischio-rectal fossa fat (m). Rectum (j). Arrows indicate access to the pudendal pedicle from above through the levator ani fibers, and from the perineum.

## Outcome measurement

The main outcome criterion was the bladder dehiscence/ prolapse rate at 3 months. Secondary outcome criteria included urinary tract infection, urethral stenosis, urethrocutaneous fistula, and parietal hernia.

This retrospective study complied with the Declaration of Helsinki principles; the parents of patients referred at birth had all the possible options explained, and a singlestage combined delayed bladder closure was performed after informed consent was obtained.

# Results

During the 27-month study period, a one-stage combined bladder closure and Kelly RSTM procedure was performed in 27 patients with bladder exstrophy: 20 had untreated bladder exstrophy (primary repair, group 1), either presenting late (n = 5) or intentionally delayed for a single-stage reconstruction (n = 15), and seven had complete bladder dehiscence (n = 6) or severe bladder prolapse (n = 1) after primary closure attempt (redo cases, group 2). There were 18 males (13 primary cases, 5 redos) and

nine females (7 primary, 2 redos). Reconstruction was performed at the median age of 3 months [0.5-37] in primary cases and 10 months [8-33] in those with previous failed attempt at bladder closure.

Antireflux procedure was performed in 22/27 cases, either with a submucosal cranial ureteral reimplant (n = 14) or a midline Gil-Vernet trigonoplasty (n = 8). After ventral transposition of the urethra, the meatus was left in hypospadias position in all boys: penoscrotal (n = 6), mid-penile (n = 10), or coronal (n = 2). None of the patient underwent posterior or innominate pelvic osteotomy. Abdominal and pelvic closure were performed with pubic approximation (n = 14) or after rectus abdominis medial transposition (n = 13).

## Outcome

Bladder closure was successful in all cases, as none of the patients had bladder dehiscence postoperatively, with a follow-up of 12 months [3–30]. No case of corpora or glans ischemic loss was observed.

Urethral fistula or stenosis occurred in a total of eight children (4 after primary repairs and 4 after redo surgery,



Fig. 2 Immobilization in a thermoformed posterior splint.

Table 1 Patients characteristics and results.									
#	Gender	Age @ surgery (mo)	Rectus abdominis transposition	Urethral fistula	Urethral stenosis	Stenosis treatment	Parietal complication	Follow-up (months)	
Prin	nary repair								
1	M	0.5	-	-	Yes	Balloon dilatation ×3	-	25	
2	Μ	7	Yes	_	_		_	18	
3	Μ	1	Yes	Yes	_		_	18	
4	Μ	24	Yes	-	-		Midline muscle dehiscence	12	
5	Μ	10	Yes	-	-		Hernia above iliopubic ramus	12	
6	Μ	3	Yes	—	—		-	14	
7	Μ	4	Yes	—	—		-	13	
8	Μ	2	Yes	Yes	_		_	10	
9	Μ	2	Yes	_	_		-	5	
10	Μ	2	-	_	_		-	6	
11	Μ	37	-	_	_			3	
12	Μ	12	-	_	_			4	
13	Μ	2	_	_	_		_	4	
14	F	4	-	_	_		-	20	
15	F	5	-	Yes	_		-	25	
16	F	3	Yes	_	_		-	12	
17	F	1	-	_	_			3	
18	F	35	_					3	
19	F	2	_	_	_			3	
20	F	2	-	—	—		-	4	
Red	o repair								
1	Μ	9	-	Yes	Yes	Balloon dilatation ×1	-	30	
2	Μ	10	_	Yes	_		_	23	
3	Μ	11	Yes	-	Yes	Balloon dilatation ×2	_	24	
4	Μ	33	Yes	-	Yes	Meatoplasty + dilatation ×3	_	13	
5	Μ	9	-		-			3	
6	F	8	Yes	-	-		_	11	
7	F	21	Yes	-	-		-	11	

Table 1). A bladder neck or urethral fistula was diagnosed in five cases (4 males, opening dorsally at the penopubic junction; 1 girl, through the mons pubis reconstruction), which resolved spontaneously in 3w to 3m in all but one case. The remaining case with persistent fistula from the bladder neck 23 months after surgery is currently awaiting repair.

Distal urethral postoperative stenosis was observed in four boys (3/5 among male redo cases, as compared with 1/ 13 of male primary cases). In two patients, it was a mild asymptomatic stenosis diagnosed on routine cystoscopy performed at 12 months postoperatively. In two cases, the stenosis was revealed by febrile urinary tract infection and/ or epididymitis. Urethral stenosis was treated successfully with 1-3 sessions of endoscopic high-pressure balloon dilatation in three patients and meatoplasty with dilatations in one case. One boy without endoscopic evidence of urethral stenosis on repeated examinations but with dilating upper-tracts was started with clean intermittent catheterization per reconstructed urethra 8 months after surgery.

In three cases, secondary vesicoureteral reimplantation was necessary at 4, 10, and 11 months after Kelly repair performed without cranial reimplantation, in a context of recurrent urinary tract infections (UTIs), absence of urethral stenosis, and persistent vesico-ureteral reflux (VUR).

Among the 13 children in whom a rectus abdominis muscle mobilization and midline transposition had been performed, one required surgical repair of a symptomatic hernia through a lateral defect above the iliopubic ramus, and one showed mild muscle dehiscence on the midline, covered with intact skin, and did not require additional parietal surgery.

# Discussion

These preliminary results suggest that bladder exstrophy closure can be safely combined with Kelly RSTM in delayed primary and even redo cases, without the need for pelvic osteotomy or demonstrable risk of bladder dehiscence.

The RSTM developed by Kelly in the 1970s was originally proposed as a staged strategy comprising neonatal closure of the bladder template, followed by the reconstruction of urethra, sphincter, and penis in males at the age of 6 months [3,7]. This procedure offers a unique opportunity of anatomical reconstruction of the internal and external sphincter muscles of the bladder neck and the urethra based on the assumption that functional structures contributing to continence do exist and need to be detached from the abnormal pelvic ring. In theory, the RSTM allows both anatomical reconstruction and tension-free soft-tissue closure, without the need for pelvic osteotomies [8].

Unlike MSRE and CPRE techniques, the Kelly procedure implies a radical detachment of the urogenital diaphragm muscles from the medial pelvic walls, through an area that is often fibrotic and scarred when performed after neonatal closure. Indeed, classical principles of bladder closure technique emphasize the need for adequate incision of the urogenital diaphragm to ensure satisfactory placement of the bladder neck and the posterior urethra deep in the pelvis [9]. Considering that the RSTM philosophy aims at precise mobilization and use of these structures for reconstruction of the continence mechanism, it seemed logical to favor a single-staged approach of untouched bladder exstrophy and so avoid the need to perform this dissection through scar tissue, in accordance of its original author view. However, the Kelly repair remains a long and technically challenging procedure even in experienced hands, with a well-established risk of erectile tissue ischemic loss [10] that could be hazardous to undertake in hemodynamically precarious newborns.

There have been an increasing number of reports in the recent years advocating a delayed approach for exstrophy

reconstruction. Historically, delayed closure was reserved for children with very small and polypoid bladder templates. Several authors now favor a delayed approach for bladder closure [6,11] or CPRE [12,13]. A delayed repair strategy offers the possibility of elective referral to an expert center, adequate establishment of mother-child bond, and may provide a better hemodynamic and metabolic window than the neonatal period in terms of healing and neurovascular risk [14]. Legitimate concerns regarding the impact of environmental exposure on the extruded bladder mucosa have been raised, especially of an increased risk of severe epithelial changes and metaplastic transformation. However, Rösch et al. showed that delayed closure did not induce detectable premalignant changes and that mucosal inflammation and cystitis cystica were not more frequent [15]. In addition, Ferrara et al. and Cervellione et al. suggested that some microscopic changes, such as squamous metaplasia, reverse to normal after bladder closure [6,16]. Other histological changes increased collagen-to-smooth-muscle such as the ratio most likely represent abnormalities at an early stage of bladder development preceding normal filling/emptying cycles with potential reversibility after successful closure [17]. In addition, there is no evidence that leaving the bladder open for a time has any adverse effect on bladder function. According to previous studies from Manchester and the Johns Hopkins Hospital, it seems that delay before bladder closure is not detrimental to bladder muscle development nor bladder growth potential with children reaching similar cystographic characteristics in the long term compared with those who undergo neonatal closure [6,18]. It appears that failure of bladder closure is actually the only independent factor resulting in less than optimal bladder growth [18].

Beyond the controversy on the role of pelvic osteotomy in the neonatal period, most high-volume exstrophy institutions recommend that delayed primary repairs or redo-closures be combined with osteotomy [1,6,13,19]. Ebert and Rosch from Regensburg however reported on a 30-year successful experience of delayed closure in children aged 6-8 weeks, without osteotomy [11]. Similarly, the London GOSH group reserves the use of pelvic osteotomy for infants older than 6 months at primary closure [20]. In the specific context of bladder closure combined with a Kelly repair, the soft-tissue mobilization itself obviates all needs for osteotomy and allows a tension-free reconstruction of the urogenital complex. Although safe closure of the abdominal wall is certainly facilitated by osteotomy in older infants, pubic approximation seems to be almost always achievable with interpubic sutures [20]. However, this maneuver has been consistently associated with an increased risk of penile ischemic injury by increasing pelvic pressure leading to compartment syndrome [14,21,22]. It has been the study authors' experience with the RSTM that despite opening the internal obturator fascia all along the Alcock's canal, extensive outward exteriorization of the corpora combined with pubic approximation may produce some kink of the neurovascular bundles, resulting in venous congestion and potential arterial spasm; hence, pubic approximation was omitted whenever possible and instead rectus abdominis mobilization was performed to ensure tension-free abdominal wall closure.

The principles of a strategy based on a combined bladder closure and Kelly RSTM is similar to the delayed CPRE and represents a paradigm shift from the classicalstaged approach for surgical reconstruction of exstrophy [23]. Assuming that delaying bladder closure is not as deleterious as previously suspected, a single-stage approach combining closure and infravesical reconstruction seems an attractive option. In addition to early bladder cycling, the RSTM in primary cases offers a unique opportunity to perform an anatomical reconstruction grounded on unscarred muscular structures. In common with the complete penile disassembly technique [24], this strategy is obviously not a true single-stage procedure because hypospadias is inevitable in most boys who will need to undergo subsequent reconstructive procedures. Adequate positioning of the bladder neck and the posterior urethra within the pelvic cavity pulls the penis backward, especially with the extreme mobility provided by a radical mobilization. Disconnection of the urethral plate from the glans permits the achievement of both adequate intrapelvic position of the vesicourethral unit and sufficient exteriorization of the penis [25]. Caione et al. described an alternative technique by lengthening the urethral plate with multiple Z-plasties [26], but it is unknown whether the length gain would be sufficient to avoid hypospadias in the Kelly repair.

Urethrocutaneous fistula is the most common complication following CPRE, occurring in 20%–40% of cases [19,27], which is similar to findings of this study. Most fistulas usually close spontaneously in a few months time after repair [28], as confirmed in the study authors' limited experience. The rate of urethral stenoses among males, although some were minor and asymptomatic, remains a significant concern in the present study series, especially in redo cases. Although it is believed that radical mobilization and hypospadias formation reduces tension in the reconstructed urethra and should not theoretically be associated with an increased risk of stenosis, it is possible that disconnection of the urethral plate from the glans jeopardizes some distal collateral blood supply and be accountable for distal stenosis.

The main limitations of the present study are the relatively small sample size and short follow-up. However, it still allows the main issue of successful closure to be addressed. Bladder dehiscence, as a failure of bladder exstrophy closure, always occurs within the first weeks after primary closure, and pelvic osteotomies have been generally viewed as mandatory in the context of delayed closures and redo-closures. Hence this preliminary report understandably focused exclusively on the risk of early bladder dehiscence and urethral complications after onestage combined closure and RSTM without osteotomies. Having the opportunity to be referred exstrophy patients after failed neonatal closures performed elsewhere, The study authors' experienced the possibility of redo-closure combined with RSTM. With the emerging concept of delayed bladder closure, this approach was extended to primary exstrophy cases, and it was believed that early RSTM combined with bladder closure in a one-stage procedure is an attractive first-line option in the lifelong management of classical bladder exstrophy.

# Author statements

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# Ethical approval

This study complied with the principles of the Declaration of Helsinki (1964) and received approval from the Institutional Ethical and Clinical Research (Centre d'Investigation Clinique) review board (Nantes, 2015).

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## Competing interests

None declared.

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