Complete Penile Disassembly for isolated epispadias repair with Mitchell-Caione technique: two cases

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Abstract. Isolated male epispadias defect is present in 10% of cases of epispadias-exstrophy complex. Many surgical technique have been described for repair of male epispadias. The aim of surgical repair is to get cosmetically acceptable and functional penis with a ventrally located urethra opening at the tip of the glans and moreover to give the patient a urinary continence. Surgical repair of epispadias malformation remains debatable as evident by the different techniques adopted. We present two cases of our experience where we used the complete penile disassembly tecnique by Mitchell-Caione in wich we acts on the voluntary control of urination by the reapproximation of the muscular plane of pelvic diaphragm and elevator muscle around the posterior urethra reconstructed. Epispadias repairs was performed on two incontinent male epispadias patient, aged 2 to 3 years. Both cases were untreated. Complete disassembly of penil components was performed to the corporal attachments down to the horizontal branches of pubic bones. The corporal bodies and the glans were split on the sagittal plane according to Grady-Mitchell. The central portion of the urethral plate was preserved intact. An electric stimulator was used to identify and reapproximate at the midline the muscular fibers that constitute the periurethral muscular complex, as a part of the anterior perineal membrane. "Z plasties" were necessary according to initial urethral length and width: the urethra was reconstructed over a double catheters, using 5/0 polyglicolic acid interrupted sutures in a single layer. In one patient we sutured symphisis pubis opened precedently, using 1/0 polyglicolic acid. The urethra was placed ventrally in the reconstructed shaft under the two corpora as Mitchell technique. Glanduloplasty was then performed. Patients were followed up for one month (June 2012). The penis of the two patients had a satisfactory cosmetic appearance with no dorsal chordee. Morover, our patients obtained urinary continence. The limitation of our study was the low follow-up. The electric stimulator used to identify pelvic muscle components in the sagittal plane for reapproximate the posterior tabularized urethra to form the periurethral muscle complex and the "Z-plasties" that elongated the urethra significantly. This procedure seems to be satisfactory for the repair of cases of isolated male epispadias because we have good results in cosmetic appearance and in urinary continence. Our study is a preliminary report and a longer follow-up with a larger number of cases is needed to document further the success of the procedure.

Key words: urethra, muscle, striated, epispadias, urinary incontinence.

INTRODUCTION

Epispadias is a rare diagnosis and most commonly described as a part of the bladder exstrophy complex. Epispadias is characterized by failure of the urethral plate to tubularize on the dorsum with defect ranging from glandular to penopubic location. In addition, male patient demonstrate a dorsal chordee whereas female patient exibit a bifid clitoris [1,2,3]. Epispadias repair is one of the great technical challenges in pediatric reconstructive urology.

Since the original description by Cantwell, multiple modifications have been made to the technique of Cantwell. As a tertiary referral center, we received two cases of epispadias and we present two cases of our experience where we used the complete penile disassembly technique by Mitchell-Caione based on a reapproximation of the muscular plane of pelvic diaphragm and elevator muscle around the posterior urethra reconstruction in order to obtain the voluntary control of the continence [4,5].

CASE 1

A 2-years child, with male epispadia of II grade, was referred to our unit. He was born with this rare anomaly. His parents were consanguineous (first cousins). Both the prenatal and perinatal history records were unremarkable.

Examination of the genitalia revealed epispadia of II grade, the scrotum was normal in size, containing normal

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descended testes. The anus was in its normal location.

Plain pelvic radiographs showed separation of the pubic rami and a normal spine. Renal ultrasonography did not demonstrate any associated renal anomaly. Cistourethrography didn't show vescical uretheral reflux (Figure 1).

The patient was treated with the technique of Mitchell-Caione. The urethra was tubularized over a 10F stent that was removed on 19th post-operative day. We followed the patient up for 5 months (from June to November). Cosmesis was good there was no sever upper tract dilatation, no renal function deterioration was observed. After five months he sensed voiding stimulus and obtained continence.

CASE 2

A 3-years child, with male epispadia of III grade was referred to our unit. He was born with this rare anomaly, his parent were first cousins, and his prenatal and perinatal history records, also, were unremarkable.

Examination of his genitalia revealed epispadia of IUII grade, with open bladder neck, a more severe form. The anus was anteriorly, the scrotum was normal in size, containing normal descended testis (Figure 2).

Plain pelvic radiography showed separation of the pubic rami and normal spine. Renal ultrasonography did not demonstrate any associated renal anomaly. Cistourethrography didn't show vescicouretheral reflux. The patient was treated with the technique of Mitchell-Caione, with the closure of the bladder neck. The urethra was tubularized over a 10F stent that was removed on 19th post-operative day. We followed the patient up for 5 months (from June to November 2012). Cosmesi was good, there was no severe upper tract dilatation, no renal function deterioration was observed, but he didn't was urinary continence.

SURGICAL TECHNIQUE

The patient underwent surgery under total anesthesia with epidural analgesia. Local adrenalina subcutaneous injection was used. The penis was surgically disassembled into three components: right and left corporeal bodies, with they respective hemiglans and the urethral wedge (i.e. urethral plate, underlying corpora spongiosa and vascular pedicle). The urethral plate is dissected from one side and elevated off the corporal body by entering the plane on the tunica albuginea of the corpora. The plate should be dissected as thick as possible to develop well-vascularized tissue. The urethral plate vascularity is based on proximal blood supply and vascularized controlateral edge. Each corpus and hemiglans are totally separated and dissected from their mates relying on separate blood supply. Separation is continued proximally with division of the attachment of the suspensory ligament to the horizontal branches of pubic bones. The pubis attachments of the corpora are left intact [6].

The corporeal bodies were then rotated internally and reapproximated with interrupted 3-0 polyglicate sutures on the dorsal surface. Because the penis was completely disassembled, internal rotation of the corpora was sufficient to correct the dorsal penile curvature. The urethra was brought to each hemiglans ventrally to create an orthotopic meatus [7]. The lenghthening of the bladder neck and urethra was accomplished with a seres of 4-6mm incision performed along the lateral aspect of the urethral plate, starting proximal to the bladder neck and ending up at the tip of the urethra (Figures 3 and 4). The central portion of the urethra plate was preserved intact. An electric stimulator was used to identify and then reapproximate at the midline the fibers that constitute the muscular periurethral complex as a part of anterior perineal membrane.

The urethra was tubularized over a 10 F stent, using







Figure 2.

5/0 polygluicole acid interrupted sutures in a single layer (Figures 5 and 6). The corporeal bodies were then rotated internally and reapproximate with interrupted 1/0 polyglicate sutures on the dorsal surface and this rotation was sufficient to correct the dorsal penile curvature.

The urethra was then brought to each hemiglas ventrally to create an orthotopic meatus, under the two corpora as in Mitchell technique. The suture line feel deep, protected by the corpora themselves. Glanduloplasty was then performed.

DISCUSSION

Many surgical techniques have been described for the repair of male epispadias. The aim of the surgical repair is to achieve a cosmetically acceptable and functional penis with a ventrally located urethra opening at the glans. Cantwell [2] initially introduced a technique based on complete mobilization of the urethral plate that was then tubularized and transplanted ventrally between the corpora. Penile disassembly techniques resulted in a real improvement in the outcomes of epispadias repair. Mitchell and others [5,8] in 1996 introduced the complete penile disassembly technique. We used Mitchell technique with Caione's modification, in this way the complete penile disassembly makes possible appropriate reassembly around the posterior urethra in more anatomical position within the pelvis with the use of electrical stimulator.

Reconstruction of epispadic urethra in the male patient is an important step of this process, with the aim not only to repair the anatomy of the external male gen-



Figure 3.

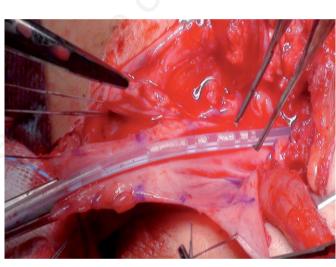


Figure 5.

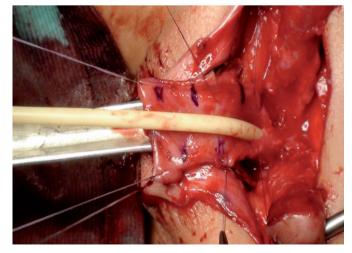


Figure 4.

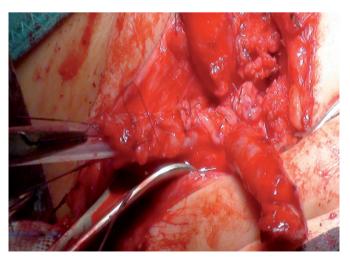


Figure 6.

italia but also to restore the structures for urinary continence [9,10]. In order to achieve capacity and continence, the main point is to restore the anatomy as close as possible to the "normal" [2,5,11]. Indeed, the elongated urethral plate allows easier deep repositioning of the urethra within the pelvic floor, restoring the angle between the ventral/horizontal male urethra, with the use of electric stimulator to identify the periurethral muscles. These strictures are usually present in patients with the exstrophy-epispadias complex but displaced more lateral than in normal children due to pelvic bone dysmorphysm. The electric stimulator enable us to observe delicate contraction inside the perineal body tissue. The dissected corpora are medially rotated and secured in position by caverno-cavernostomy. The urethral plate is tabularized to the ventral side of the rotated corpora.

We used this technique because we believed that the complete penile disassembly with the use of electrical stimulator can provide normalization of the urthra and penis, in fact it makes possible appropriate reassembly around the posterior urethra in a more anatomical position within the pelvis. In normal human anatomy and physiology the urethral sphinteric mechanism has an intrinsic and extrinsic or periurethral component. The intrinsic urethral sphincter has a smooth muscle component with thin inner longitudinal and circular smooth coats [12] surrounded by striated external component, the so-called urethral rhabdosphincter, wich is sleeve-shaped from bladder neck. In male the striated external urethral sphincter is a tubular cylinder of muscle extending from the perineal membrane to cover the whole membranous urethra [13]. It extends proximally over the prostate as an attenuated layer and is part of the anterior fibromuscular stroma of the prostatic urethra. The external striated urethral sphincter or rhabdosphincter is innervated by the pudendal and pelvic nerves [14,15]. Connective structures of the periurethral sphincter complex that support the urethra arise from the back of the pubic bones and insert into the perineal body in the midline and into the urethral striated muscle sphincter [15,16]. Urethral closure in maintained at rest by mucosal sphincter mechanism and low energy muscle activity. Urethral closure is reinforced by increased smooth and striated urethral muscle tone. The pubovisceral or periurethral muscular complex, mostly the puboperineus and pubovaginalis muscles, that insert on either side into the perineal body as structural components of the pelvic floor slings. Thus, voluntary urinary control under stress and the forward progression of seemen into the anterior urethra are ensured [12, 17].

The advantage of penile disassembly lies in the potential for better correction of the chordee, along with penile lengthening [9]. The main disadvantages of the Mitchell technique are that complete urethral plate dissection is usually followed by shortening and retraction, and the risk of urethral plate devascularization, especially in its distal part, is always present [9,18]. Numerous modification have been applied to prevent the occurrence of hypospadias, including suturing the urethral plate with interrupted sutures, augmenting the urethral plate using preputial or parameatal skin flaps, or modification of the total penile disassembly technique using either cavernostomy or limited penile disassembly to achieve a glandular meatus [19,15]. In the presented technique, the main point is the creation of multiple "Z-plasties" wich start proximal to the veru montanum. As a consequence, we are able to create a funnel shaped bladder neck, longer and narrower than it would be without the "Z-plasties", and to elongate [20].

The limitations of our study are that the follow-up is not so longer so we needed to extend it.

CONCLUSIONS

Epispadias reconstruction using the Mitchell-Caione repair can achieve total or near complete urinary continence, decreasing the morbidity of multiple procedures, and potentially allowing children to achieve continence at an earlier age. The penis of the two patients had a satisfactory cosmetic appearance with no dorsal chordee. Morover, our patients obtained urinary continence. The limitation of our study was the low follow up. The electric stimulator was important to identify pelvic muscle components in the sagittal plane for reapproximate the posterior tabularized urethra to form the periurethral muscle complex and the "Z-plasties" that elongated the urethra significantly. This procedure seems to be satisfactory for the repair of cases of isolated male epispadias because we have good results in cosmetic appearance and in urinary continence. Our study is a preliminary report and a longer follow-up with a larger number of cases is needed to document further the success of the procedure.

REFERENCES

- 1. Frimberger D. Diagnosis and management of epispadias. Semin Pediatr Surg 2011;20:85-90.
- 2. Cantwell FV. Operative treatment of epispadias by transplantation of the urethra. Ann Surg 1895;22:689-94.
- 3. Ransley PG, Duffy PG, Wollin M. Bladder exstrophy closure and epispadias repair. In: Spitz L, Nixon HH, eds Rob and Smith's operative surgery pediatric surgery. Boston: Butterworths; 1988. pp 620-622.
- Gearhart JP, Sciortino C, Ben-Chaim J, et al. The Cantwell-Ransley epispadias repair in extrophy and epispadias: lessons learned. Urology 1995;46:92-5.
- 5. Mitchell ME, Bagli DJ. Complete penile disassembly for epispadias repair: the Mitchell technique. J.Urol 1996;155:300-4.
- 6. Ibrahim M, Mohamed Y, Hazem RI, Hosam H. Partial penile disassembly for isolated epispadias repair. Pediatr Urol 2008;71:235-8.
- 7. Ashraf TH, Tamer H. Complete Penile Disassembly for epispadias

repair in postpubertal patients. Pediatr Urol 2011;78:1407-10.

- Kibar Y, Roth C, Frimberger D, Bradley PK. Long term results of penile disassembly technique for correction of epispadias. Pediatr Urol 2009;73:510-4.
- 9. Zaontz MR, Steckler RE, Shortliffe LM, et al. Multicenter experience with the Mitchell technique for epispadias repair. J Urol 1998; 160:172-6.
- 10. Grady RW, Mitchell ME. Management of epispadias. Urol Clin North Am 2002;29:349-60.
- 11. Braga LH, Lorenzo AJ, Bagli DJ, et al. Outcome analysis of isolated male epispadias: single center experience with 33 cases. J Urol 2008;179:1107-12.
- 12. Lawson JON. A functional anatomy of the lower urinary tract in children. Pelvic anatomy of the lower urinary tract in children. Pelvic anatomy III. Contemp Issue Pediatr Urol 1995;9:57.
- 13. Kaye KW, Creed KE. Anatomy and innervation of the external urethral sphincter. Dial Pediatr Urol 1997;20:3.

- 14. Hollabaugh RS Jr, Dmochowski RR, Steiner MS. Neuroanatomy of the male rhabdosphincter. Urology 1997;49:426.
- 15. Carroll PR, Dixon CM. Surgical anatomy of the male and female urethra. Urol Clin North Am 1992;19:339.
- 16. Pena A. Surgical management of anorectal malformation:a unified concept. Pediatr Surg Int 1998;3:82.
- 17. Caione P, Capozza N, Lais A, Matarazzo E. Periurethral muscle complex reassembly for exstrophy-epispadias repair. J Urol 2000; 164:2062-6.
- 18. Hammouda HM. Results of complete penile disassembly for epispadias repair in 42 patient. J Urol 2003;170:1963-5.
- 19. Hafez AT, El-Sherbiny MT. Complete repair of bladder exstrophy: management of resultant hypospadias. J Urol 2005;173:958-61.
- 20. Caione P, Nappo S, Matarazzo E, et al. Penile repair in epispadiasexstrophy complex: can we prevent resultant hypospadias? J Urol 2013;189:1061-5.

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