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 exhibit 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 in which 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. Moreover, 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 exhibit 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...
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 vesicourethral 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 IUII 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 vesicourethral 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). Cosmesis 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 adrenaline 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 lengthening of the bladder neck and urethra was accomplished with a series 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
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-

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**Figure 3.**

**Figure 4.**

**Figure 5.**

**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 stricture 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 mediially rotated and secured in position by caverno-cavernosotomy. 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 urethra 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 sphincteric 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 external urethral sphincter is a tabular cylinder of muscle surrounding the posterior urethra in a more anatomical position within the pelvis. 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 epispidias 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

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