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REVIEW

Congenital megaprepuce: review of the literature

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Congenital megaprepuce (CMP) is a specific cause of buried penis which occurs as an isolated anomaly. The cause of CMP is not known. However, developmental disruption has been confirmed histologically by the presence of dysplastic dartos tissue. Many surgical techniques have been described, but none seems to be better than another in all aspects.

In this narrative review of the literature, we aim to provide a comprehensive, critical and objective analysis of the current knowledge on the topic.

Keywords: congenital megaprepuce, buried penis, dysplastic dartos tissue

Introduction

The concept of buried penis was introduced by Keyes in 1919, when he described the inconspicuous penis. He described it as an apparent absence of the penis which exists when the penis lacks its proper sheath of skin, and lies buried beneath the integument of the abdomen, thigh or scrotum.[1,2] Many authors put forward their own terms to describe the buried and inconspicuous penis. These included concealed penis, hidden penis, trapped penis and webbed penis.[1,2] The first description of congenital megaprepuce (CMP) as a cause of buried penis was by O'Brien et al. in 1974 who described it as a buried penis characterised by ballooning of the excessively redundant prepuce that provided a ready receptacle of urine on micturition.[3]

CMP is considered a congenital penile malformation.[4] It is a specific clinical condition that results in a buried penis. The condition characteristically appears as a foreshortened penis with a wide dome-shaped base that exhibits hemispherical ballooning during micturition.[5] The swelling often subsides spontaneously, or the parent may report manual expression to expel the urine. A milky or cloudy appearance and/or an unpleasant smell may characterise the urine. Although a non-retractile and ballooning foreskin with micturition is considered normal in infancy, the magnitude of swelling in CMP is characteristic.[3,6] In their description of the first case of CMP, O'Brien et al.[3] noted the excessively redundant inner prepuce which they called a preputial bladder. The presence or absence of true phimosis is not agreed upon in the literature. Alexander et al.[5] noted the presence of an unusually proximal and stenotic preputial opening leading to ballooning as urine flows against the resistance. With time the inner prepuce preferentially stretches and expands beneath the narrowed opening, forcing the glans and corpora into the pubic fat pad and upper scrotum causing dissociation of shaft skin from Buck's fascia.[5] Ferro et al. described a flap-valve effect of the anterior and posterior lips of the foreskin causing an obstruction to urine flow and no true phimosis.[7] Powis and Capps described preputial intussusception as the cause of obstruction to urine flow, which when left to progress leads to a capacious inner prepuce; hence, they also referred to the condition as acquired megaprepuce.[8]

Epidemiology of CMP

The incidence of CMP is unknown. The series described by various authors in the literature have a wide range, from as little as three cases to as many as 65 cases. The time periods during which these cases were recorded is also varied, and most studies included inconspicuous penis cases secondary to other causes and not CMP alone. Werner et al. recorded three cases over a one-year period, while Murakami et al. reported a series of 65 cases over a 14-year period.[9,10] Furthermore, the determination of the incidence is not an easy task as there is confusion regarding the definition of a buried penis and there is no universal criterion of what constitutes congenital megaprepuce. Patient presentation is often later in infancy due to parental concern about the inconspicuous penis, the swelling during micturition and the need to manually express urine out of the capacious inner prepuce.[5,9,11,12] A total of 868 cases was reported in literature until 2011 but these cases had different inclusion criteria.[12,13]

Aetiology and pathology of CMP

The cause of CMP is largely unclear. A noticeable aspect is the unretractable, redundant inner prepuce.[4] Hadidi noted contrasting observations by different authors in their description of the morphology of a buried penis.[12] A distinct fibromuscular layer tethering the penile shaft to the anterior abdominal wall was described by Crawford while Wollin et al. emphasised the abnormal skin mobility over the penile shaft skin.[14,15] Also, in his series of 60 patients, Hadidi[12] noted that the abnormal mobility of penile shaft skin and fascia was a constant finding. Abnormal attachments of the fusiform ligament and the suspensory ligament were found in 32 patients in Hadidi's series.[12] Liu et al. found dysgenetic fusiform ligament attached to distal or middle penile shaft in all 22 cases over a five-year period. Redman did not, however, find abnormal fascial attachments in his series of 31 patients who were managed for congenital buried penis.[16,17]

Embryologically, CMP is thought to develop following failure of separation of migrational planes in the developing male external genitalia.[18] The genital tubercle that is located at the cranial end of the urethral folds rapidly elongates to form the phallus pulling forward the urethral folds which then fuse over the urethral plate to form the penile urethra. Concurrently, the scrotal swellings that initially develop in the groin, migrate caudally and medially to unite across the scrotal septum. Tethering of the penile corpora to the deep fascia with a high scrotum results in failure of separation of these developmental planes.[19,20] Histological examination of dartos fascia resected in patients with congenital penile pathology including congenital buried penis and hypospadias, showed abnormal dartos tissue. Spinoit et al. confirmed, histologically, poorly developed hypotrophic smooth muscle fibres and the randomly distributed smooth muscle fibres with no parallel configuration in dartos tissue resected in patients with congenital buried penis and hypospadias.[21]

Presentation

The classical presentation of CMP is severe pooling of urine in the large redundant preputial reservoir during voiding.[5,22,23] Some degree of discomfort is reported as crying, abnormal facial expressions during voiding or awakening before passing urine.[19] The ballooning of the penile base is often described as egg, orange or cricket ball size.[3,24,25] The swelling subsides spontaneously as the urine dribbles out slowly, or often the parent reports manual expression of the urine out of the foreskin.[5] The urine is described as cloudy and/or foul smelling. Cases that presented with a urinary tract infection have been reported in the literature; however, this is a rare presentation.[5,22,26]

On examination, the hemispherical swelling is noticeable especially after voiding. This subsides with manual expression. A perceived normal size phallus and glans can be felt under the abdominal or scrotal skin on palpation.[5,23] An ill-defined penoscrotal angle with some penoscrotal transposition is often observed.[22,27] No other associated congenital abnormalities are present. However, Hirsch et al. reported a degree of hypospadias with chordee in six patients with congenital megaprepuce.[27]

CMP and classification of buried penis

Buried penis, concealed penis, hidden penis and engulfed penis are non-specific umbrella terms that have been used to refer to the inconspicuous penis. The more commonly used term is buried penis. However, one can get confused when surgical techniques are described as these techniques are not applicable to every case of buried penis. Accuracy is important when describing the condition causing the buried penis appearance.[19] Various classification systems have been described in the literature. A classification system that highlights the cause of the inconspicuous penis and helps in the accurate anatomical description of each pathological lesion that may lead to its appearance has been proposed by Alexander et al. Generally, there are about four causes of a buried penis.[5] All are characterised by a normal-sized phallus.[5,19,24] The first is trapped penis due to distal preputial scarring following circumcision. Summerton et al. point out that this is due to a missed diagnosis of CMP managed by standard circumcision.[24] The second is buried penis due to excessive suprapubic fat and poor fixation of penile shaft skin. Campbell described this in 1951 and used the term concealed penis.[28] Maizels et al. described the same condition as buried penis secondary to suprapubic adiposity.[1] The third is buried penis secondary to congenital megaprepuce. The purpose of this review and the described novel surgical repair technique is specific to this clinical entity. The presentation and the findings on examination under anaesthesia for CMP are classic. The fourth is webbed penis. Maizels et al. did not classify this as buried penis.[1] While it is a cause of inconspicuous penile appearance, there is no buried penis appearance. Alexander et al. also puts webbed penis as a cause of inconspicuous penis but not buried.[5]

Indications for surgery

The indications for surgery are both functional and cosmetic improvement. Parents are anxious about the inconspicuous appearance of the penis and the swelling that develops during micturition as a result of urine accumulating in the foreskin. These micturition troubles of ballooning of foreskin, need for manual expression, episodes of balanitis, occasional urinary tract infections, urinary retention and spraying of the urinary stream are common indications for surgery. Liu et al. reported dorsal curvature and severe penile skin shortage as the main indications for surgical correction in their series of 22 patients with congenital completely buried penis.[16]

Surgical management techniques

A multitude of surgical repair techniques have been described for the management of buried penis. The techniques, however, were not quite specific to buried penis secondary congenital megaprepuce. Of note is that the different surgical techniques obey similar surgical principles. The differences between these techniques are basically on how penile shaft coverage is achieved. Shalaby and Cascio broadly categorise these into single-stage and two-stage approaches.[19] The staged repair relieves the initial obstruction of urine flow and discomfort from the narrowed preputial ring. This is achieved by performing a dorsal or ventral or two lateral slit incisions at the constricting ring which allows remodelling of the inner preputial skin to take place, and the need for further surgery can be assessed at the 4–6 years follow-up.[19] Unpublished data by Tasker et al. noted that this is often satisfactory in certain cultures where traditional circumcision is required as a right to passage.[29] The single stage techniques provide penile shaft coverage with either outer shaft skin, or inner preputial skin, or a combination of the two.

Anatomical approach

The anatomical approach technique was developed by Cuckow in 1998 and published in 2000.[30] It is a single-stage repair technique that utilises outer skin for penile shaft coverage. The initial step is identifying the line of demarcation between penile shaft skin and scrotal skin ventrally. A curved or smiling face incision is made along the line followed by dissection between dartos fascia and Buck's fascia to free the penis from its deep tethering and caudal mobilisation of the scrotum. Dissection is continued distally lifting

the preputial sac off the penile skin. The preputial sac is opened ventrally, followed by excision of the redundant inner preputial layer leaving a sub-coronal cuff approximately 6–8 mm wide. The created dorsal rectangle or quadrilateral flap of penile skin is thinned by excising excess dartos tissue. Laterally the flap is wrapped around the shaft on both sides and sutured distally to the mucosal cuff. Closure of the ventrally placed now diamond shaped opening is done vertically or longitudinally recreating the median raphe before closing the wound with a pressure dressing.[18,19,31] Dorsal preputial flaps can be used for shaft coverage as described by Podesta et al.[26] The final appearance is that of a circumcised penis.[18] Similarities have been observed between this method of repair and other authors, including modifications to the anatomical approach. These include:

a. **Transverse scrotal incision** by Joseph where a transverse scrotal incision is made and extensive dissection at the root of the penis is done.[32] Low fusion of the penile corpora was thought to be responsible for the buried penis.[32] Sub-dermally placed fine sutures are used to anchor the skin to the tunica to avoid re-tenting of the skin that leads to recurrence of the penile concealment. Penile shaft coverage is achieved using penile skin only and the final appearance is that of a circumcised penis.

b. **Omega scrotal incision** by Leao et al. and modified by Hirsch et al. [22,27] The technique consists of an omega shaped incision made at the penoscrotal junction. This is followed by caudal mobilisation of the scrotum which leads to some gain in penile length. A circumferential incision of the inner prepuce 5 mm from the coronal sulcus is made and excess redundant inner prepuce excised. The dermis of the penile shaft skin is then sutured to Buck's fascia at the base of the penis ventrally on either side of the corpus spongiosum to recreate the penoscrotal angle. Skin of the phimotic ring is preserved and used for penile shaft coverage. A dorsal incision is made for a few millimetres to release the tension. The incision corrects the penoscrotal transposition and the anchoring sutures at the base recreate the penoscrotal angles and prevent recurrence of the penile concealment.

Unfurling method

Use of unfurled prepuce to cover the shaft in buried penis repair was first described by Donahoe and Keating in 1986.[33] This is a single stage repair that utilises inner preputial skin. The redundant inner preputial skin rapidly acquires the appearances similar to that of the remaining foreskin.[33] Modifications to this technique were made by Shenoy and Rance as they included some tailoring, where excision of a v-shaped wedge of ventral skin was done followed by approximation of the edges longitudinally to recreate the midline raphe.[2,5]

Fixing, unfurling and tailoring

This procedure was described by Ruiz et al. in 2011. It is a modification with improvement of the procedure originally described by Donahoe and Keating in 1986, and revised by Sheno and Rance in 1999.[2,33,34]The first step is the release of the phimotic ring by a limited resection of the phimotic ring to deliver the glans. Dissection is then done in the soft tissue plane between prepuce and Buck's fascia all the way to scrotum ventrally and dorsally continued to the suspensory ligament. The redundant inner prepuce is then unfurled with minimal to no dissection to avoid damage to the lymphatics. Tailoring is done to reduce the redundant inner prepuce and, ventrally, a triangle of inner prepuce is removed in the midline to permit resurfacing of the penile shaft. The proximal outer preputial skin is fixed to Buck's fascia and ventral suturing done in the midline to recreate the median raphe.[34] Furthermore, Rod et al. described variations to this technique with an initial dorsal incision to unfurl the whole inner prepuce then a ventral longitudinal incision.[35] The whole penis is freed by subcutaneous dissection off Buck's fascia. Recreation of the penoscrotal and penopubic angles are done by suturing the dermis of the penile skin to Buck's fascia ventrally and dorsally avoiding the corpus spongiosum and the neurovascular bundles, respectively. Lastly, the inner prepuce is reduced ventrally by excising and tailoring of two triangles with their subcutaneous tissue. This reduces the risk of oedema.[35]

Single-stage repairs with combined penile and inner preputial skin

The DOLOMITE

This technique was described by Callewaert et al.[23] It consists of two longitudinal incisions. The first incision is made on the ventral side down to the scrotum, followed by unfurling of the inner prepuce on penile shaft skin. Then a dorsal longitudinal incision is made on the unfurled prepuce up to the sub-coronal level. The third and fourth incisions are made on each side of the shaft connecting the most proximal part of the ventral incision to the dorsal midline incision. Finally, the fifth and sixth incisions begin from the sub-coronal level dorsally and run in an oblique direction across the inner prepuce to the penoscrotal junction one on each side. The dartos is freed off the Buck's fascia and 2–4 quilting sutures are placed between skin and Buck's fascia to re-establish the penoscrotal and penopubic angles at the base of the penis.[23]

Ventral V-plasty

Described by Alexander et al., the first step is stretching the preputial opening using mosquito forceps to deliver the glans, then further cleaning the glans and inner preputial layer.[5] This is followed by a ventrally placed incision in the midline to divide the tight constricting band. Unfurling of the inner prepuce is done and placed proximally towards the base of the penis. Dorsally, an incision is made on the inner prepuce leaving 5 mm of sub-coronal collar and ventrally extended into a V-shape. Degloving of the penile shaft is done and redundant dartos fascia completely resected with bipolar cautery to ensure good haemostasis. Anchoring sutures are done at the base of the penis at 5, 7 and 12 o'clock positions to recreate the penoscrotal and penopubic junctions. The V-shaped inner preputial flap is then fashioned into the opposite ventral V-shaped defect in the penile skin.[5]

Arc incision surgical approach

Described by Lin et al.[4] as a single-stage repair, this procedure commences with an arc incision on the ventral prepuce, followed

by progressive eversion of the redundant inner prepuce. Then, 5 mm from the coronal sulcus, a circumferential incision is made, followed by a longitudinal ventral incision linking the two incisions. Penile degloving is done exposing Buck's fascia. Resection of the unfurled inner preputial layer is then done, carefully preserving blood supply to the outer layer, which would be used for shaft coverage. The penopubic angle was reconstructed by fixing the proximal dorsal outer prepuce to Buck's fascia at the base, avoiding the neurovascular bundles. Dissection and resection of the subcutaneous tissue around the base was done through a small transverse incision at the penoscrotal junction. Outer prepuce from the body of the penis was moved to the frenulum and one from the base to the ventral side. A new penoscrotal junction was created with suturing between Buck's fascia and the dermis. The rest of the suturing was done and scars were confined to circular sub-coronal and longitudinal ventrally.[4]

Penoplasty techniques

Initially described by Brisson et al. in 2001, Perger et al. re-described this single stage penoplasty technique for repair of buried penis in infants and children.[36,37] The procedure begins with a circumferential incision 2–3 mm from the coronal sulcus, followed by a vertical incision ventrally to the base of the penis. Dissection is done between dartos and Buck's fascia and complete degloving of the penis is done to the pubic rami dorsally and penoscrotal junction ventrally. All tethering bands are released leaving the suspensory ligament intact. A fan-shaped flap is developed by unfurling the preputium with help of silk sutures placed on the edges. The penile shaft is then secured to the prepubic fascia at three sites, just distal to the junction of the corporal bodies avoiding the neurovascular bundles, for penile lengthening and to prevent retraction. Skin is attached to the base of the penis on multiple sites circumferentially. The ventral incision is closed longitudinally and the sub-coronal sutures done circumferentially at the coronal sulcus.[36,37]

Chuang[38] described a penoplasty technique at the Chang Gung Memorial Hospital in Kaohsiung, Taiwan in 1995. The notable difference was the initial dorsal incision to deliver the glans. The incision was then extended distally to an unspecified distance from the sub-coronal sulcus and finished off circumferentially. Penile degloving was done to the base and anchoring stitches placed between skin and penile shaft to immobilise the basal foreskin. The rest of the foreskin is then spread, and an adequate length of ventral prepuce preserved. The halves of the foreskin flaps are transposed to the dorsal side and approximated in a Z-plasty fashion to aid in shaft coverage.[38]

Hadidi[12] also described an operative technique similar to some extent to the penoplasty technique by Brisson et al. and Perger et al.[36,37] Hadidi started with placing two stay sutures ventrally to stretch the prepuce.[12] A ventral midline incision was made from the tip of the prepuce down to the penoscrotal junction to deliver the glans. Glans stay suture and urethral catheter were inserted. Dissection to base was done followed by measurements of the inner prepuce length and distance from base to tip of the glans. The decision to divide the suspensory ligament was made based on whether the penis retracted inside the pubis or not after stretching, which corresponded to a grade 2 buried penis according to his classification. Excess suprapubic fat was noted and resected in grade 3 buried penis as it reduced the effect of dividing the suspensory ligaments. Fixation of the tunica albuginea to the perostium at symphysis pubis and pubic bones laterally, was done followed by fixing penile skin at the base to tunica albuginea. Excess long inner prepuce was excised before circumferential sub-coronal suturing to the mucosal collar leaving a circumcised appearance of the penis.[12]

Liu et al. described a penoplasty technique that includes penoscrotal Z-plasty to gain skin length for shaft coverage.[16] Their study was based on 22 children aged between 2.5 and 5.8 years. They used Crawford's 1977 classification of buried penis and only recruited patients with the complete type. The procedure started with a circumferential incision on the narrow outer foreskin, then a ventral cut along the midline down to the scrotum. The skin and tunica dartos are completely dissected off of Buck's fascia and all adhesions or chordee are resected ventrally into the penoscrotal junction. Dissection on the dorsal aspect is then carried down to the base of the penis near the pubic bones to expose the fundiform ligament. This was attached to the distal or middle shaft of the penis. Release of the fundiform ligament was done in all cases taking precaution not to damage the dorsal neurovascular bundle. Recurrent penile retraction was prevented by fixation sutures between the dartos layer and lateral tunica albuginea at the base of the shaft. Penoscrotal Z-plasty is then made to increase skin length for shaft coverage. The inner prepuce is cut off leaving a 5 mm collar. Reapproximating of the inner and outer foreskin, and the median raphe, are then done. Finally a compression dressing and foley catheter are placed, which are removed on day three after the operation.[16]

Cromie et al. also described an anatomical alignment technique for the surgical correction of buried penis.[39] Their series had 74 patients. The procedure started with the placement of a stay suture on the glans penis for traction. This is followed by a circumferential incision leaving a 1 cm sub-coronal mucosal collar. Release of constricting band vertically with minimal to no shaft skin involvement then follows. Penile degloving to the base of the penis is done followed by unfurling of the redundant foreskin if needed for shaft coverage. Dermal sutures are placed on 10 and 12 o'clock positions at the base, and if necessary on the middle shaft too, to ensure good fixation of skin to Buck's fascia. The preputial collar and shaft skin are realigned and sutured in a fashion similar to that for a standard circumcision.[39]

Use of flaps in buried penis repair

In 1995, Boemers and De Jong described a surgical technique for correction of buried penis that used preputial flaps.[40] Their technique started with release of the phimotic ring followed by placement of a stay suture on the glans. A sub-coronal incision is made leaving 3 mm of sub-coronal collar. Degloving of the penis is then done taking care not to damage the neurovascular bundle. Then the prepuce is unfurled preserving its blood supply. This stage creates a more or less rhomboid shaped skin flap. These flaps are then

used to accomplish penile shaft coverage in one of two ways. In the ventral-lateral technique, the distal preputial margin is sutured to the coronal sulcus starting dorsally in the midline. Then the two lateral tips are brought down into the upper scrotum. Alternatively, the crossing over technique can be used. This consists of crossing the lateral tips of the rhombic shaped prepuce anteriorly, pulling one tip down laterally to the base of the penis and the other one up sub-coronally to the opposite side. Anchoring the tunica dartos to Buck's fascia at the base and along the shaft, laterally, is done to prevent retraction of the penis. A compression dressing is applied over the penis postoperatively and a transurethral catheter inserted. Both are removed on day three after the operation.[40]

Kojima et al. described the use of preputial island pedicled flaps to correct buried penis in congenital megaprepuce.[41] In this procedure the first step was release of the phimotic ring with a 3 cm long ventral incision along the midline. The glans is then delivered followed by placing a traction stay suture on the glans. A sub-coronal circumferential incision is made and another one at the junction of inner and outer foreskin. The outer preputial skin is then dissected off the shaft at the level of the dartos fascia down to the penopubic junction. The inner preputial skin is dissected off the shaft between dartos and Buck's fascia. This creates an island pedicled flap of inner prepuce with dartos fascia. A window is then made in the base of the pedicle for buttonhole trans-positioning of the island flap to the ventral side of the shaft. Dartos fascia is sutured circumferentially at the base to prevent penile retraction. The flap is circumferentially wrapped around the ventral side of the penis and re-sutured on to itself on the dorsum. Any excess skin is excised and finally the inner preputial skin covers the dorsal and proximal shaft of the penis.[41]

Delgado-Miguel et al. described the use of dorsal dartos flaps in the correction on buried penis secondary to congenital megaprepuce.[11]

In 2020, Alsmahy et al. described the use of Byar's flaps in the management of buried penis secondary to congenital megaprepuce.[42] The procedure is started with a small ventral midline slit on the tight preputial ring to enable retraction of the foreskin. After fully retracting and unfurling the prepuce, a circumferential incision is made 5 mm from the coronal sulcus, then complete penile degloving is done up to the penopubic and penoscrotal junctions. Excess redundant inner prepuce is then excised together with bulky dartos fascia. The penoscrotal and penopubic angles are then recreated by placing anchoring sutures between skin and Buck's fascia sparing the neurovascular bundles. Byar's flaps are then prepared and rotated ventrally before they are sutured in place in a similar fashion to Byar's stage 1 urethroplasty. Lastly, circumferential suturing of the mucosal collar is done and a tight compression dressing is applied.[42]

Complications of buried penis repair surgery

There is varied incidence of surgical complications after buried penis repair according to different authors. Alexander et al. and Murakami et al. recorded zero complications and zero recurrences in their series of 10 and 65 patients, respectively. [5,10] Betancor et al., however, recorded severe oedema and wound infection in their series of 61 patients.[31] In general, complications of CMP or buried penis repairs include haematoma formation, urinary retention, wound infection and breakdown, preputial necrosis, severe protracted oedema, and in worst case scenarios dorsal chordee and recurrence of the penile concealment, may occur. Reoperations have been recorded less frequently. Some surgical techniques are associated with complications. Procedures that have transverse incision at the base of the penis and procedures that utilise inner preputial skin for penile shaft coverage, have been noted to have severe prolonged oedema postoperatively.

Conflict of interest

The authors declare no conflict of interest.

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Ethical approval

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