African Urology 2024;4:16-20
https://doi.org/10.36303/AUJ.0111

AFRICAN UROLOGY
ISSN 2710-2750 EISSN 2710-2750

ORIGINAL RESEARCH

Congenital megaprepuce: description of a simple, reproducible novel surgical technique

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Background and objectives: A buried penis, secondary to congenital megaprepuce (CMP), is a specific clinical entity plagued by the non-uniform use of terminology and many surgical repair techniques that have brought confusion to this subject. The results of surgery have been mixed and no technique is superior to another. This study aims to provide a brief review of the literature on CMP, describe a simple, reproducible novel surgical repair technique, and retrospectively review the first series of patients managed by this method.

Method: A total of 16 boys aged between 10 and 43 months were referred with CMP and operated on by the same surgeon (JH) from 1 February 2017 to 31 July 2022, using the same method, termed an "inverted circumcision".

Operative approach: See Appendix A: Video of inverted circumcision procedure. A ventral longitudinal incision crossing the penoscrotal junction was made on all patients. Dissection was done between the dartos fascia and Buck's fascia. The penis was delivered through the incision for the inverted circumcision. Anchoring sutures were placed between the dermis and Buck's fascia to recreate the penoscrotal and penopubic junctions. The phallus was replaced into the shaft skin and the excess inner prepuce was trimmed accordingly. Circumferential suturing was done and all patients had the appearance of a circumcised penis. The minimum follow-up was six weeks.

Results: Good to excellent anatomical outcome was subjectively recorded according to assessment by the reviewing surgeon. None of the patients needed reoperation to improve the outcome.

Conclusion: The inverted circumcision technique is a simple, reproducible technique and should be added to the surgical options of correcting a buried penis secondary to CMP.

Keywords: congenital megaprepuce, reproducible novel surgical technique

Introduction

A multitude of surgical techniques used to repair CMP have been described. Each has its advantages and disadvantages; however, most have common steps that have become principles of buried penis repair. None is better than another and there is no gold standard repair technique. The main differences emanate from whether it is a one- or two-stage repair and the source of skin for penile shaft coverage. Some use penile shaft skin while others utilise the inner preputial skin for shaft coverage.[1-3] The different methods described in the literature were not specific to the management of CMP but buried penis/inconspicuous penis in general.

The appropriate age at which surgical correction should be undertaken is not agreed on in the literature. Perger et al. mentioned that the condition may self-resolve with sexual maturation.[1] Summerton et al. highlighted that the condition is obvious at birth, or shortly thereafter, and recommend surgical intervention as soon as the diagnosis is made as there was no evidence of spontaneous resolution with growth and development.[2,3] Routine circumcision is contraindicated in cases of congenital megaprepuce. This would remove the skin that will be required for shaft skin coverage. The condition (trapped penis) can be a result of routine circumcision in patients with congenital megaprepuce.[3]

This paper aims to describe a novel, and what we consider a reproducible, surgical technique for the repair of CMP and describe the outcomes in this initial series of patients.

Materials and methods

Ethical and institutional approval was obtained (HREC 638/2022). An urology operations database was accessed and searched for patients who were operated on for CMP. The search words were: buried penis, CMP repair, modified circumcision, and inverted circumcision. The search was done from 1 February 2017 to 31 July 2022. Folder numbers were obtained and folder requests from hospital records were made. A comprehensive folder review was done, and data were obtained and recorded on an anonymised data sheet.

The information collected was the age at diagnosis, age at surgery in months, and the reason for presentation regarding symptoms and signs such as inconspicuous penis, swelling of foreskin during micturition, need for manual expression of urine, dripping of offensive urine, perceived painful micturition, and urinary tract infection (UTI). The examination findings recorded included the presence of hemispherical swelling if the glans was palpable in the foreskin or not, retractability of the foreskin, suprapubic fat pad, expression of urine from the foreskin, and subjective comments on the penis shaft size as small or normal. Records of the postoperative complications, such as swelling, wound infection, wound breakdown, the final appearance of the penis, the need for redo-surgery, and also the final comment on cosmesis, were obtained and recorded into a data collection sheet.

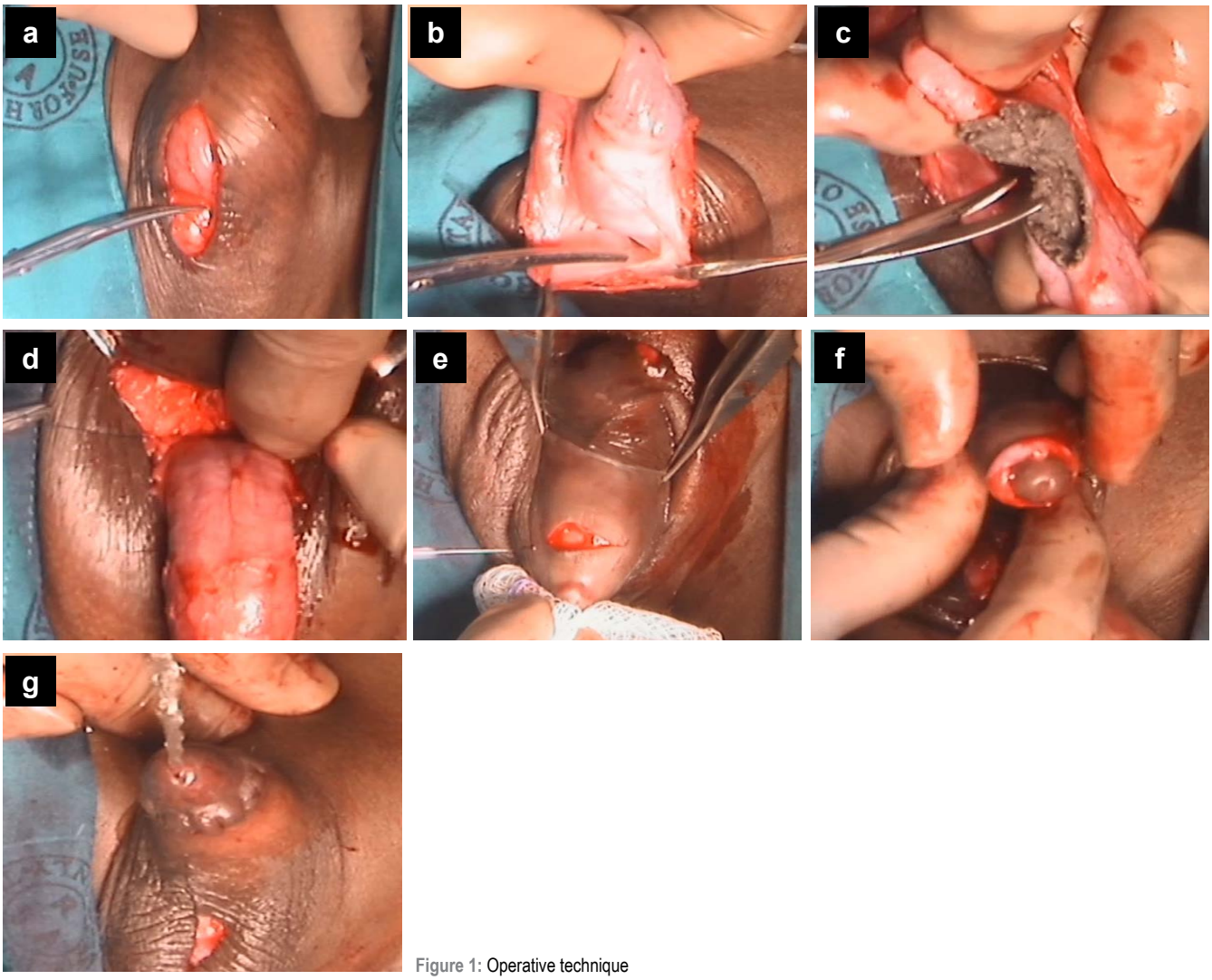

Figure 1: Operative technique

Operative technique

See Appendix A: Video of inverted circumcision operative technique. The operations were performed under general anaesthesia by the same surgeon (JH). A stat dose of cefazolin antibiotic was given to all patients before the skin incision was made. The first stage was a ventrally placed longitudinal incision over the penoscrotal junction (Figure 1a). This was followed by a careful dissection between the dartos fascia and Buck's fascia of the penis (Figure 1b). This dissection was carried out on either side, as well as proximally and distally, to free the penis from the abnormally bulky dartos fascia. The penile shaft was then delivered through the incision and further dissection was distally made (Figure 1c).

The next step is the inverted circumcision, using a pair of scissors and protecting the glans penis (Figure 1d). This is followed by the placement of anchoring sutures at both the penoscrotal and penopubic junctions to recreate the penoscrotal and penopubic angles, taking care not to injure the urethra and the neurovascular bundles (Figure 1e). Excess subcoronal mucosal collar was excised if necessary to 3–5 mm wide (Figure 1f). The penis was then replaced in the skin sheath and circumferential suturing was done like standard circumcision. The ventral incision was closed, and the final, immediate appearance postoperatively was that of a circumcised penis (Figure 1g).

Results

A total of 19 folder cases were found and reviewed. Out of the 19 folders, only 16 cases met the selection criteria. Their diagnosis was a buried penis secondary to CMP and were operated on during the period stated using the novel repair technique (inverted circumcision). Three of the cases did not meet the selection criteria. Two of the three patients had a modified circumcision for the removal of dorsal hooded foreskin in glanular hypospadias and one patient had routine circumcision for an unspecified reason.

The ages at diagnosis ranged from 5 to 41 months and the age at surgery ranged from 10 to 43 months. Six patients were referred from the day hospitals, five from private general practitioners, four as self-referral, and one from the medical outpatient clinic.

The clinical presentation and examination findings are shown in Table I below. Inconspicuous penis and swelling of the foreskin with micturition were the predominant presenting complaints, while the most common examination findings were hemispherical swelling, expression of urine on applying pressure, and a non-retractable

foreskin. Only one patient had confirmed increased suprapubic fat on examination together with hemispherical swelling.

Table I: Presentation and exam findings frequency table

Presenting complaint and examination finding	*n* = 16
Inconspicuous penis	13
Swelling of the foreskin during micturition	13
Manual expression of urine	9
Dripping of offensive urine	8
Perceived painful micturition	5
UTI	3
Confirmed non-palpable glans within the foreskin	6
Confirmed hemispherical swelling	10
Urine expressed on applying pressure	7
Confirmed non-retractable foreskin	8
Confirmed normal penile size	6
Confirmed increased suprapubic fat	1

Results of the postoperative complications, final appearance, need for redo-surgery, and final comments on cosmesis were recorded. No patient developed wound infection and/or breakdown. Penile swelling was short-lived and subsided spontaneously. All patients had the final appearance of a circumcised penis. Neither recurrence nor scarring was noted. None of the patients required redo-surgery after at least six weeks of follow-up. Final comments of the surgeon's subjective appraisal of cosmesis were made in 9/16 cases and were recorded as well healed in two, good in five, very good in one, and excellent in one.

Discussion

CMP is now a well-recognised cause of congenital buried penis. The history goes as far back as 1919 when Keyes described a buried penis, then in 1974 when Gwinn et al. presented a case of unusual presentation of phimosis. However, much of the credit goes to O'Brien et al. who first described excessive preputial ballooning on micturition to constitute a preputial bladder and termed it CMP.[4,9]

Rod et al. clearly stated that CMP is a specific form of buried penis.[5] Callewaert et al. mentioned that CMP is obvious from birth and tends to worsen within the first few months of life.[6] The condition is rarely missed as it is eye-catching to the parent or caregiver, as well as the medical personnel. In a series of six patients over five years, Callewaert et al. noted that penile malformation was evident from the early months of life.[6]

The most common description of CMP noted in the literature is the great redundancy of the inner preputial skin and a normal penile shaft and glans. Also noted as part of the clinical description is a non-retractable foreskin that balloons with micturition. These descriptions were noted in all the patients in our series. The failure to retract the foreskin is attributed to the narrowing of the preputial opening and abnormal bands of dartos tissue attached to the distal shaft of the penis. Also, the ballooning noted on micturition is attributed to phimosis.

The authors of this article do not agree with Alexander et al. who pointed out that phimosis is present in all cases of CMP.[7] We noted that a flap valve effect of the ventral lip of the foreskin over the dorsal lip created resistance to urine flow as well as accumulation in the foreskin during micturition. As the urine fills within the inner prepuce, it tends to push down the penile shaft and glans, such that without surgical correction the condition worsens. Powis and Capps described a similar appearance and called it preputial intussusception.[8]

Limited literature is available to explain the abnormality. The early presentation and deficiency of skin ventrally support an anomaly of development. Repeatedly mentioned by different authors is the failed separation of migration planes of the penis during development. This results in reduced ventral skin, dysplastic dartos tissue, and poor anchorage of the skin at the base of the penis. We agree with Hadidi et al. and Werner et al. that abnormal bands of dysplastic dartos attach to the distal shaft of the penis, holding the phallus within the integument of the abdominal or scrotal skin.[9,10]

The exact incidence of CMP is not known and is difficult to identify. The condition is relatively rare and each manuscript reports a small number of cases. Shalaby et al. pointed out that patients typically present between 3 and 18 months of age. In our series, the age at presentation ranged from 5 to 41 months with 17 months as the average.[11]

The most common presentation in our series was ballooning of the foreskin with micturition, inconspicuous penis, and the need for manual expression to empty urine (87.50%, 81.25%, and 62.50%, respectively). This affirms what is documented in the literature. Rod et al. suggested that ballooning may be associated with a degree of discomfort to explain the finding by other authors about awakening from sleep and facial expressions during voiding attributed to the pain and discomfort.[5] In our series, perceived pain on micturition was documented in 5/16 cases. It is a subjective presenting complaint. Equally uncommon is confirmed febrile UTI. Only three cases had UTI in our series. This finding confirms the trend in the literature that CMP rarely presents with a UTI. More common is urine with an offensive smell but not proven UTI. The smell is most likely due to the stasis of urine in the megaprepuce.

On examination, the most common clinical finding was hemispherical ballooning, noted in 10/16 patients, and urine could be expressed in 7/10 patients with swelling. The glans penis was not palpable within the foreskin in six patients, and the foreskin was non-retractable in 8/16 patients. Of the remaining patients, there was no comment on whether the glans was palpable in the foreskin or whether retraction was possible or not. This highlights the fact that the assessment of each patient upon physical examination is not uniform among the practitioners.

The finding of an ill-defined penoscrotal angle with some magnitude of penoscrotal transposition can be noted on examination. However, the presence of other congenital malformations is rare. Surprisingly, Hirsch et al. reported a degree of hypospadias in six of their patients with CMP, a finding that has not been confirmed by other researchers.[12] We agree with Shalaby et al. that CMP is an isolated condition not associated with hypospadias.[11]

Surgical repair of CMP is recommended as soon as the diagnosis is made.[7,8,13,14] The waiting period in our patient series ranged from one to eight months. All cases were booked electively on the next available date. There is no documentation of spontaneous resolution of CMP with age in the literature. As suggested by many authors (Alexander et al., Leao et al., Shenoy et al., and Powis and Capps) the hemispherical swelling tends to worsen with age.[7,13,15,16] Surgical correction is required to restore normalcy. Unfortunately, the suggestion or recommendation by Powis and Capps that standard circumcision is all that is required to correct the anomaly is misleading.[16] Healing with scaring over the glans resulting in a trapped penis has been noted after a standard circumcision for CMP.

Surgical correction of CMP requires observing certain steps in the repair. Many surgical techniques have been described, but not all of them are specific to CMP. We add our repair method to the array of alternative techniques. This method is a single-stage repair technique that achieves shaft coverage using outer skin and leaves only a rim of subcoronal inner prepuce, just like a standard circumcision. It observes the important steps in the repair of CMP, which are the excision of excess inner prepuce, excision of excess redundant dartos tissue, and reconstruction of both the penoscrotal and penopubic angles; careful not to injure the urethra ventrally and the neurovascular bundles dorsally.

The advantages of this novel repair method are that it has less post operation oedema because it leaves a narrow band of submucosal collar and has no transverse incisions at the base of the penis that disrupts the lymphatic drainage of the penile skin. It achieves skin coverage with outer preputial skin, which gives a better cosmesis compared to methods that utilise inner preputial skin. Again, the suture lines are limited to the ventral aspect over the median raphe and circumferentially as in standard circumcision suturing.

The potential disadvantages of this technique are the limited exposure and haematoma formation. Adequate exposure is ensured by making a long enough incision across the penoscrotal junction to allow for easy dissection and access to the penopubic space for placement of anchoring sutures to recreate the penopubic angle. The risk of haematoma formation is abated by ensuring adequate haemostasis using bipolar cautery as one resects the redundant dartos tissue. None of the 16 patients in this series had haematoma formation as a postsurgical complication. The postoperative oedema resolved spontaneously by the sixth week postoperatively.

Reoperation surgery is reported in the literature with different repair methods. In our series with this novel repair technique, no cases needed reoperation. Also, on review of the folders, none of the patients had a second operation to correct a complication or persistence of CMP. All cases had the final appearance of a circumcised penis with no scarring.

This study is, however, limited by the fact that it is a retrospective study and the sample size is small. The follow-up period was also short, ranging from six weeks to three months. Possible complications or need for repeat surgery may be required as both the sample size and duration of follow-up are increased.

We recommend an outcome analysis study to evaluate both the patient and parent satisfaction with the cosmetic appearance and functional outcome. A larger sample size and longer follow-up period would be ideal to establish long-term complications and rates of redo-surgery after this novel repair technique.

Conclusion

CMP is a specific cause of a buried penis. It occurs mostly as an isolated anomaly. The cause 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 seem to be better than others in all aspects. The described novel inverted circumcision technique is simple, reproducible and easy to learn as a repair method.

Conflict of interest

The authors declare no conflict of interest.

Funding source

Manatsa Chimhamhiwa was supported by fellowship funding from IVUmed (USA) and Kind Cuts for Kids (Australia).

Ethical approval

Research ethics and institutional approval were obtained from the University of Cape Town Human Research Ethics Committee (HREC 638/2022). Informed consent was obtained from the parents of the case used for videography.

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Video available online at https://africanurology.com/index.php/au/article/view/111/127

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