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Megameatus intact prepuce with distal megalourethral dilatation: A case report and review of the literature

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Abstract

Introduction: Megameatus Intact Prepuce is a rare and often under-recognised variant of distal hypospadias, as the normal-appearing foreskin may mask the underlying urethral abnormality.
Case Presentation: We report the case of a male neonate who presented with voiding difficulty and preputial ballooning, in whom Megameatus Intact Prepuce was identified during an attempted circumcision. The child subsequently underwent hypospadias repair at eight months of age. Intraoperative findings revealed a subcoronal megameatus with associated distal megaurethral dilatation extending to the mid-penis. Redundant urethral tissue was excised, and tubularised plate urethroplasty was performed with a satisfactory outcome.
Conclusion: This report highlights the anatomical variability of Megameatus Intact Prepuce and the importance of careful clinical assessment and tailored surgical management to achieve favourable outcomes.

Keywords: Foreskin; Hypospadias; Infant; Penis/abnormalities; Penis/surgery

Introduction

Hypospadias is a congenital abnormality resulting from incomplete embryogenesis of the penis, leading to variable anatomical abnormalities including a ventrally displaced urethral opening, ventral penile curvature, and a dorsally hooded prepuce with ventral preputial deficiency.[1] It is the second most common congenital anomaly in males after undescended testis.[2] Megameatus Intact Prepuce (MIP), first described by Duckett and Keating in 1989, is a rare distal variant of hypospadias, occurring in approximately 1 in 10,000 male children and accounting for 1–3% of all hypospadias cases.[3-5] Unlike more common forms, MIP is characterised by a wide, patulous meatus and a wide, deeply grooved urethral plate, all concealed beneath an entirely intact prepuce and typically occurring in the absence of penile curvature.[6]

The normal appearance of the prepuce means that MIP is frequently overlooked at birth and often recognised only during routine neonatal circumcision or evaluation of voiding difficulties.[4] Although MIP has been described globally, published reports from sub-Saharan Africa remain sparse, and awareness among clinicians is limited.[7] This case highlights practical lessons in the management of this concealed hypospadias variant and adds to the limited regional literature by describing our operative approach and outcome.

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Case presentation

A 2-day-old term male neonate, was delivered via spontaneous vaginal delivery after an uneventful perinatal course. The mother's antenatal history was unremarkable, and there was no family history of congenital penile anomalies. He was referred to our unit because of difficulty in passing urine, associated

with ballooning of the prepuce and dribbling during micturition.

On examination, he was an apparently healthy neonate with stable vital signs. Genital examination revealed an intact prepuce with a stenosed preputial opening. During voiding, there was distal ballooning of the prepuce, and gentle pressure expressed urine. Palpation demonstrated a ventral groove. There was no evidence of chordee or penile torsion. The scrotum was well formed, with both testes palpable within the scrotal sacs.

Baseline haematological and biochemical investigations were normal. Abdominal ultrasonography demonstrated no abnormalities. A silicone urethral catheter was inserted to ensure urinary drainage.

On the tenth day of life, an attempt was made to perform circumcision to relieve the preputial narrowing and urinary difficulty. Retraction of the prepuce at that time revealed a wide urethral meatus consistent with Megameatus Intact Prepuce, and circumcision was deferred pending definitive repair.

At eight months of age, after satisfactory growth and optimisation, the patient underwent tubularised urethral plate urethroplasty under general anaesthesia. Intraoperative findings included a subcoronal megameatus with ventral redundancy of the distal penile skin (Figure 1) and a dilated megaurethra extending to the mid-penile shaft.

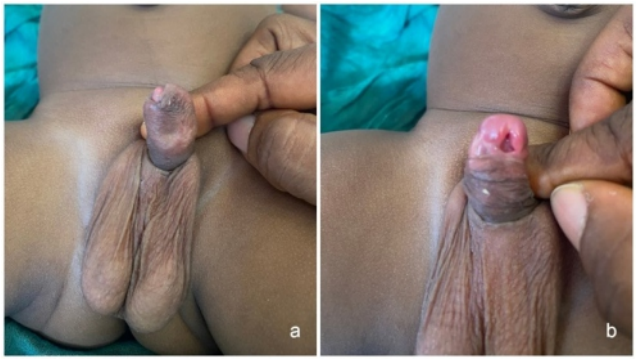

Figure 1 Preoperative views of the penis: (a) intact prepuce with ventral redundancy of the distal penile skin, and (b) retracted prepuce demonstrating a wide subcoronal urethral meatus.

The penis was degloved through a circumferential incision 5 mm below the corona, with dissection extended to the penoscrotal junction. The ventral aspect was dissected superficially in the plane between the megaurethra, dartos tissue, and skin. The glans wings and mucosal collar were mobilised, and the megaurethra was incised vertically up to the mid-penile shaft, where the normal urethra began. The width of the urethral plate required for urethroplasty was measured, and the redundant tissue excised bilaterally. The urethra was tubularised over an 8-Fr silicone catheter in two layers using 6-0 vicryl (Figure 2a), with a spongioplasty forming the second layer. The suture line was covered with a vascularised dartos flap, followed by glanuloplasty and skin closure (Figure 2b). The urethral catheter was left in situ as a stent.

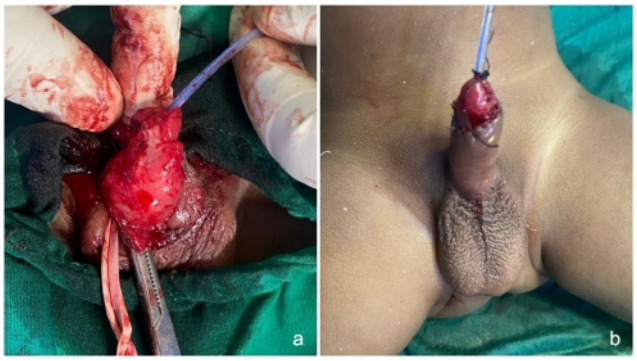

Figure 2 Intraoperative views: (a) tubularisation of the urethral plate over a silicone catheter, and (b) completed urethroplasty following glanuloplasty and skin closure.

Figure 3 Postoperative view three weeks after surgery.

The postoperative course was uneventful. The catheter was removed on day 14, and the child voided with a strong urinary stream. At follow-up, he had an acceptable cosmetic appearance with mild penile torsion, but no evidence of fistula, meatal stenosis or urethral stricture (Figure 3). He has been scheduled for elective correction of penile torsion at a later date.

Discussion

Although hypospadias is a common penile anomaly, Megameatus Intact Prepuce (MIP) is rare and may evade diagnosis because the external appearance of the penis may be deceptively normal. In our patient, the diagnosis became evident only during an attempted circumcision undertaken to relieve preputial narrowing and urinary dribbling. Despite published descriptions of this anomaly, awareness among primary care workers may be limited. As a result, many boys with MIP may remain undiagnosed or may not be referred promptly for specialist evaluation.[5,8] To the best of our knowledge, only one other case from sub-Saharan Africa has been reported in the literature, suggesting that MIP may be under-recognised within the region.[7]

MIP demonstrates a spectrum of anatomical variations, ranging from minor widening of the meatus to more complex forms with deep glanular grooves and distal urethral dilatation.[6,9] Our patient presents an uncommon variant with megaurethral dilatation extending to the mid-penile shaft. This finding is not frequently documented, however, it lies within the recognised morphological continuum of MIP. Distal urethral ballooning or megalourethra is believed to result from inadequate development of the corpus spongiosum.[10] This often leads to functional disturbances, which in our patient included dribbling and preputial ballooning during voiding. The thin, dysplastic and redundant urethral plate, together with spongiosal deficiency, typically require excision of excess urethral tissue followed by tubularisation with multilayered closure.

Differentiating MIP from other distal urethral anomalies is important for selecting the most appropriate operative strategy. Anterior urethral valves and urethral diverticula may present with distal ballooning but usually appear as a ventral saccular outpouching proximal to the meatus, typically between the bulbous and mid-penile urethra, without the broad meatus and glanular defects characteristic of MIP.[11] Similarly, congenital megalourethra may involve urethral dilatation but lacks the meatal and glanular features seen in MIP.[10] Distal hypospadias with dorsal hooding can be distinguished by its incomplete preputial development and common association with ventral chordee.[2]

A range of operative techniques has been described for the management of MIP, and the choice depends

Table 1 Summary of selected published case series and reports on Megameatus Intact Prepuce, highlighting anatomical findings, operative approaches and postoperative outcomes.

Author & Year	Country	Number of patients	Key Findings (n)	Surgical Technique(s)	Outcomes
Bhat et al., 2017.[14]	India	12	Classical distal MIP comprising glanular (2), subcoronal (6) and distal penile (4) variants	TUPU in 11, frenuloplasty alone in 1; preputial preservation with frenuloplasty in 10	No complications, good cosmetic and functional outcomes
Cendron, 2018.[9]	United States	32 (25 operated, 7 observed)	Distal MIP with wide meatus, deep glanular groove and intact prepuce, 7 had minor glanular dilation	TUPU in 10, Mathieu repair in 15	Overall good cosmetic and functional outcomes, Glans injury in 1 and urethrocutaneous fistula in 1. Better outcomes noticed with the Mathieu technique
Musa et al., 2019.[7]	Nigeria	1	Circumcised child with markedly wide urethral meatus (6 mm)	GAP	Good cosmetic and functional outcome; no complication
Duan et al., 2020.[4]	China	27	Distal MIP comprising glanular (2), coronal (15) and distal penile (10) variants	TIPU in 13; TUPU in 7; Mathieu in 5; GAP in 1; MAGPI in 1	Overall complication rate 14.8% (mainly Urethrocutaneous fistula in two, meatal stenosis in two). No clear superiority of one technique
Ekberli et al., 2020.[13]	Turkey	31	Distal MIP with frequent median raphe anomalies (10); other associated anomalies including UPJO (2), bilateral undescended testes (1), chordee (2) and penoscrotal web (1)	TIPU in 5; TUPU in 16; Meatoplasty in 10	Good cosmetic and functional outcomes with urethrocutaneous fistula in one patient.
Permana et al., 2021.[16]	Indonesia	1	Distal MIP with subcoronal meatus	TIPU	Good cosmetic and functional outcome; no complication
Hakimi et al., 2023.[5]	Afghanistan	2	Distal MIP: one circumcised before diagnosis	TIPU	Good cosmetic and functional outcome; no complication.
Ramaswamy et al., 2024.[6]	Saudi Arabi	12	Noticed wide anatomical variability among patients (particularly with meatal width and urethral plate width); proposed umbrella term "Hypospadias with Intact Prepuce (HIP)" with MIP as a major subgroup as not all patients were found to have megameatus.	TUPU in 7; TIPU in 5	Good cosmetic and functional outcomes; urethral injury in 2 patients and ventral glans necrosis in 1 patient.
Prakash et al., 2024.[17]	India	1	Mid-penile meatus with chordee in a child with intact prepuce.	TIPU	Good cosmetic and functional outcome; no complication.

MIP = Megameatus Intact Prepuce; TIPU = Tubularised Incised Plate urethroplasty; TUPU = Tubularised Plate Urethroplasty; GAP = Glans Approximation Procedure; MAGPI = Meatal Advancement and Glanduloplasty incorporated; UPJO = Ureteropelvic junction obstruction.

largely on the experience of the surgeon, the meatal location, the urethral plate morphology and the glans anatomy. These include the Glans Approximation Procedure (GAP), the pyramid repair, the Mathieu perimeatal flap, and tubularisation of the urethral plate using techniques such as Tubularised Incised Plate Urethroplasty (TIPU) and Tubularised Plate Urethroplasty (TUPU).[3,4,6,12,13] The pyramid repair was introduced specifically for MIP and targets its characteristic anatomy. Glanular defects have been managed successfully using Meatal Advancement and Glanduloplasty (MAGPI) or the GAP, whereas coronal variants may be repaired with GAP, TIPU and the Mathieu repair.[4,7,9,13] Many children have a wide urethral plate that lends itself well to TUPU (or Thiersch–Duplay) repair as a suitable option. It has been noted, however, that techniques which work well for non-megameatal hypospadias may not always produce satisfactory results in MIP.[14] Bhat et al. observed that the GAP may be limited by a disparity in calibre between the neourethra and the normal urethra due to inadequate mobilisation of the glans wings, and by overlapping glanular suture lines without an intervening tissue layer, which may increase the risk of fistula formation. To address these concerns, modifications incorporating formal glans wing mobilisation and interposition of a dorsal dartos flap have been proposed to achieve a tension-free repair with improved outcomes.

The anatomical features of MIP can make reconstruction technically difficult. Dissection of the wide meatus and broad urethral plate may result in thin and fragile glans wings, which are more susceptible to dehiscence and urethrocutaneous fistula formation.[14] While TIPU allows preservation of the lateral aspects of the urethral plate and enables controlled mobilisation of the glans, this technique is most appropriate when the plate is narrow. In contrast, the broad urethral plate that characterises most MIP variants usually favours tubularised plate urethroplasty without incision. A further challenge arises in children who have already been circumcised, which reduces available penile skin and dartos tissue for coverage. Some authors have observed that circumcision does not adversely influence outcomes.[5,15] To reduce the risk of complications, multilayer repair with vascularised tissue support is widely recommended in both TIPU and TUPU. In our patient, the presence of megalourethra was associated with thin glans wings and lateral segments of the urethral plate. Excision of the redundant outer plate and tubularisation using TUPU with vascularised dartos flap support was therefore considered the most appropriate surgical approach.

Outcomes following MIP surgery are generally favourable, with reported complication rates lower than those for other forms of hypospadias.[4,6,13,14] Several series describe low rates of urethrocutaneous fistula and meatal stenosis, together with excellent functional and cosmetic results. Most children achieve a straight urinary stream and a well-shaped glans. Our patient's early postoperative outcome is consistent with these findings, with acceptable cosmesis and no evidence of fistula, meatal stenosis or stricture at follow-up. A summary of nine published reports is presented in Table 1. As noted earlier, based on our literature search, only one previous case has been documented from sub-Saharan Africa,[7] which highlights the need for greater awareness and improved reporting of MIP and its outcomes within the region.

Conclusion

Megameatus Intact Prepuce remains an uncommon and often under-recognised variant of distal hypospadias. Careful clinical assessment is required to distinguish it from other distal urethral anomalies and to guide appropriate surgical planning. Our case illustrates the anatomical variability that may be encountered, including associated megalourethra, and the need to tailor repair to individual morphology. Successful reconstruction is achievable with meticulous technique and adequate tissue support, and postoperative outcomes are generally favourable. The scarcity of published cases from sub-Saharan Africa emphasises the need for increased awareness and documentation of this condition to improve early diagnosis and optimise management in our region.

Reporting Guidelines: This manuscript was prepared in accordance with the SCARE guidelines for case reports.[18]

Ethical Considerations: This manuscript was prepared in accordance with the principles of the Declaration of Helsinki on research involving human subjects. In accordance with institutional policy, approval from the institutional ethics review board was not required for the publication of an individual case report. Written informed consent was obtained from the patient's parent for publication of anonymised clinical information and accompanying

images.

Conflicts of Interest: The authors declare no conflicts of interest.

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