Tubularized incised plate urethroplasty for megameatus intact prepuce hypospadias variant: First reported case in Indonesia

Wien Permana, Tarmono Djojodimedjo, Johan Renaldo
Department of Urology, Faculty of Medicine, Universitas Airlangga-Dr. Soetomo General Academic Hospital, Surabaya, Indonesia

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ABSTRACT
INTRODUCTION AND IMPORTANCE: Megameatus intact Prepuce (MIP) is a rare variant of hypospadias characterized by a wide meatus with a normally conformed prepuce, no chordee, and usually no effects in micturition or sexual physiology. However, quality of life and psychosexuality may be affected.

CASE PRESENTATION: A 6-year-old-boy was referred by a general practitioner to the hospital due to an abnormality of the urethral meatus. The patient did not have any complaints. A large external urethral orifice at the ventral area of the coronal glans was discovered. The prepuce was normally-conformed, and there weren’t any signs of chordee. The patient was diagnosed with an MIP hypospadias variant and underwent a tubularized incised plate (TIP) urethroplasty. During the follow-up, one week after the procedure, the patient did not have any complaints. Physical examination showed excellent anatomical and functional outcome based on the cosmetic appearance resembling a normal penis and urinary stream.

CLINICAL DISCUSSION: Awareness regarding this abnormality is necessary as MIP is often not diagnosed until circumcision. Even though MIP patients are usually asymptomatic, surgery should still be considered based on aesthetic and psychosexual reasons. TIP urethroplasty was chosen for this patient based on the clinical findings of this patient. TIP procedure for MIP patients could result in an excellent penile performance and function with a very low complication rate.

CONCLUSION: TIP urethroplasty is able to fulfil satisfactory cosmetic and functional outcomes for the MIP hypospadias variant. Awareness of the disorder should be increased to prevent potential psychosexual disturbances.

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1. Introduction and importance
Megameatus intact Prepuce (MIP) is a rare variant of hypospadias with an incidence of 1 in 10,000 cases accounting for 1–3% of all hypospadias cases [1]. It is characterized by a wide meatus, located on either the corona or sub corona, with a normally conformed prepuce and no chordee. Micturition or sexual physiology is usually not affected in patients with MIP [2]. However, an untreated case may potentially affect a hypospadias patient’s quality of life due to penile aesthetic and psychosexual reasons [3]. Due to the asymptomatic nature of some cases, the patients are usually diagnosed during circumcision or late in life in non-circumcised children at the time of prepuce retraction [4]. This is highly important in countries like Indonesia, where circumcision is considered a common tradition based on religious and cultural reasons [5]. The case has never been previously reported in Indonesia, even though a recently published nationwide multicenter study showed many hypospadias cases in the country [6]. In addition to the encounter rarity, reconstruction of MIP is challenging as there is spongiosum deficiency resulting in a thin and immobile ventral portion of the urethral meatus tissue [7]. A suitable surgical method is specifically designed to take the urethral opening’s shape and position, width of the urethral plate, and depth of the urethral groove to fulfill satisfactory results [1]. We report a 6-year-old male patient with a subcoronal MIP hypospadias variant successfully treated by tubularized incised plate (TIP) urethroplasty. This case report has been reported in line with the SCARE criteria [8].

2. Case presentation
A 6-year-old-boy was referred to our Hospital by a general practitioner due to an abnormality of the urethral meatus, located at the bottom surface of the penis and wider in size, found during a circumcision. Before the circumcision, the patient and his family had never noticed any irregularities or complaints. The patient’s family denied any family history of a similar condition. A large external urethral orifice at the coronal glans’ ventral area was discovered during the physical examination. The prepuce was normally-conformed. There weren’t any signs of chordee or other genital abnormalities, as shown in Fig. 1. The patient was diagnosed...
Fig. 1. The patient’s external genitalia showing an intact and normally-conformed prepuce with no chordee.

with a MIP hypospadias variant based on the physical examination findings. The pediatric urologist of our hospital planned a TIP urethroplasty for the patient. Before the procedure, the patient underwent a complete pre-operative workup to ensure a successful surgery, resulting in normal physical and laboratory results. The patient did not have any previous history of surgery or allergies. The procedure was started by placing sutures through the glans to facilitate traction. Subcoronal incision and degloving were performed to the proximal part of the penis to facilitate urethral plate release. An erectile function test was performed by injecting 0.9% saline solution through a winged-needle.

Fig. 2 shows subcoronal megameatus with intact prepuce and without a chordee. The glans wing was then separated from the urethral plate allowing mobility of the glans wing during neourethral fusion, as shown in Fig. 3. An 8 Fr. Silicone catheter was inserted, and the urethral plate edges were gently picked up and wrapped around the catheter for a tension test. We used a 6-0 Polydioxanone continuous suture for the urethral plate tubularization. Interposition of dartos flaps obtained from the dorsal prepuce layer was performed afterward. The flaps were then placed between the neourethra. Glansplasty was performed next by placing a 6-0 Polydioxanone suture at the corona, tightening the glans wings as shown in Fig. 4. The ventral skin of the shaft is tightened proximally to distally.

Excess of the skin of both sides was excised. Wound closure is finished with interrupted 6-0 Polydioxanone sutures and dressing, as shown in Fig. 5. After the procedure, the patient’s vital signs and urine production were observed. On the 5th day, shown in Fig. 6, the dressing was removed to evaluate postoperative healing. On the 7th day, the urethral catheter was removed. During the follow-up in the outpatient clinic, one week after the procedure, the patient did not have any complaints. Physical examination during the follow-up, 2 months after the patient was discharged, showed excellent anatomical and functional outcome based on the cosmetic appearance resembling a normal penis and normal urinary stream. The patient had been scheduled to come to the hospital a year after the operation; however, the patient could not come to the hospital due to financial and geographical constraints. Nevertheless, the patient and his family reported satisfactory cosmetic penile appearance and urinary function via a telephone call a year after the operation. Uroflowmetry or cystography was not performed as the patient did not have any urinary complaints.

3. Clinical discussion

There is an increasing awareness of MIP as a rare hypospadias variant [9]. As a country with more than a quarter-billion people and 600 ethnicities with a rising report of hypospadias cases, this unique variant’s report is highly necessary [6]. East Java, where the
patient currently lives, is the second-largest province in Indonesia with a wide variety of socio-demographic backgrounds representative of the country [10]. Based on religious and cultural reasons, circumcision is a daily occurrence for local general practitioners [5]. Thus, awareness and knowledge regarding this abnormality are necessary among urologists, and general practitioners as MIP is often not diagnosed until circumcision [7]. Currently, there are controversies regarding whether to operate MIP patients or not [9]. However, even though MIP patients are often asymptomatic, surgery should still be considered based on aesthetic and psychosexual reasons [11]. Several previous studies discovered that men with hypospadias tend to be more inhibited to initiate contact with the opposite sex [12]. One of the main reasons is that several patients were afraid to be mocked when naked [13]. A study by Tack et al. claimed that a vast majority of men in their study are satisfied with their childhood hypospadias repair. Most men in the study were pleased with their penile appearance even though it could be perceived as abnormal by a physician. Therefore, in deciding whether to perform surgery or not, accepting a suboptimal outcome as a surgeon is more important for the patient’s psychosexual well-being [3]. In the age of increased cosmetic awareness and concern, surgical correction is justified [4]. Currently, there is no single urethroplasty method that could provide a universal solution for every patient [1]. A technique resulting in excellent outcomes in hypospadias patients does not always translate well to MIP variants. In non-megameatal distal hypospadias, meatal advancement and glanuloplasty incorporated (MAGPI) and perimeatal-based flap techniques are usually preferable; however, they are not suitable for several MIP cases [9]. MAGPI technique is generally chosen for patients with a normal ventral urethral wall, which is not too thin and mobile [14]. Glans approximation procedure (GAP) is generally used for a MIP hypospadias variant with a wide and deep glanular groove or a wide meatus. Even though GAP is relatively easy to perform, acquiring the gland’s conical shape is difficult as the proximal gland is very small [9]. In this patient, TIP urethroplasty, as illustrated in Fig. 7, was chosen as the procedure of choice. Every patient’s procedure should be based on the location of the meatus and specific anatomic features [2]. TIP procedure could result in an excellent penile performance and a very low complication rate [15]. The technique is usually used for less than 1 cm urethral plates and patients with minimal to no chordee [16]. In this patient, favorable clinical results were seen during the two weeks post-operative follow-up. Even though several studies suggested uroflowmetry examination during the follow-up, the significance of this recommendation in patients without clinical signs or symptoms remains unclear [17]. Urinary complications may occur years after initial reconstruction. Therefore a long-term follow-up is necessary [18]. Long term follow-up of hypospadias repair is usually mandatory to identify possible late complications as many studies have discovered that primary hypospadias repair complications may present
late [19]. Even though it is of substantial importance, there is a significant lack of long-term data regarding hypospadias repair from centers and studies. Current studies have follow-up rates between 12.7%, 22.4%, and 56.2%; however, most studies do not report those lost to follow-up [17]. The patient in this report had also become lost to follow-up due to financial and geographical constraints. We were able to contact the patient’s family one year after the operation via a telephone call, by which they reported no further complaints since the last follow-up. This interaction method is supported by a pilot study that concluded that telemedicine could be utilized in pediatric urology patients’ postoperative care [20].

4. Conclusion

TIP urethroplasty is able to fulfill adequate cosmetic and functional outcomes for the MIP hypospadias variant. As a rare variant of hypospadias, awareness for the disorder should be increased to ensure proper management preventing potential psychosexual disturbances for the patient in the future.

**Declaration of Competing Interest**

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**Ethical approval**

The ethical clearance for this research was issued by The Hospital Research Ethics Committee of “Rumah Sakit Umum Daerah Dr. Soetomo” where the patient was admitted (Ethical Clearance number: 0224/129/IX/2020).

**Consent**

Written informed consent was obtained from the patient’s parent on behalf of the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

**Author contribution**

- Wien Permana: Conceptualization, Investigation, Resources, Writing – Original Draft, Supervision, Project Administration.
- Tarmono Djojodimedjo: Investigation, Resources, Writing – Original Draft, Review and Editing.
- Johan Renaldo: Investigation, Resources, Writing – Original Draft, Review and Editing.

**Registration of research studies**

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References

[1] S. Duan, X. Jiang, X. Zhang, et al., Megameatus intact prepuce treated with urethral plate-preserving surgery: a retrospective study of an unusual hypospadias variant, Transl. Androl. Urol. 8 (2019) 583.

[2] B. Aydogdu, M.H. Okur, H. Zeytun, et al., Megameatus with intact prepuce (MIP) hypospadias variant applied tubularized incised plate (TIP) urethroplasty: a rare case report, Pediatr. Urol. Case Rep. 2 (2015) 15–19.

[3] L.J.W. Tack, A. Springer, S. Riedl, et al., Psychosexual outcome, sexual function, and long-term satisfaction of adolescent and young adult men after childhood hypospadias repair, J. Sex. Med. 17 (September (9)) (2020) 1665–1675.

[4] M. Cendron, The megameatus, intact prepuce variant of hypospadias: use of the inframensal vascularized flap for surgical correction, Front. Pediatr. 6 (2018) 55.

[5] M.I. Tanggok, Circumcision law in christianity and islam, AHKAM J. Ilmu. Syariah 18 (2016).

[6] G.W.K. Duarsa, P.M.W. Tirayasa, B. Daryanto, et al., Risk factors for urethrocutaneous fistula following hypospadias repair surgery in Indonesia, J. Pediatr. Urol. 16 (2020), http://dx.doi.org/10.1016/j.purol.2020.04.011, Epub ahead of print 2020.

[7] M.A. Faasse, E.V. Dray, E.Y. Cheng, Repair of megameatus: a modified approach, J. Pediatr. Urol. 11 (2015) 100–101.

[8] R.A. Agha, T. Franchi, C. Sohрабi, et al., The SCARE 2020 guideline: updating consensus Surgical Case REport (SCARE) guidelines, Int. J. Surg. 94 (2020) 226–230.

[9] A. Bhat, M. Bhat, A. Bhat, et al., Results of tubularized urethral plate urethroplasty in Megameatus Intact Prepuce, Indian J. Urol. IJU J. Urol. Soc. India 33 (2017) 315.

[10] Y.P. Kloping, F.R. Muharram, A.M. Reswari, Validity and reliability of the Indonesian version of the international index of erectile function, J. Clin. Urol. (2020), http://dx.doi.org/10.1177/2051415820927819, Epub ahead of print 2020.

[11] B. Haid, T. Becker, M. Koen, et al., Penile appearance after hypospadias correction from a parent’s point of view: comparison of the hypospadias objective penile evaluation score and parents penile perception score, J. Pediatr. Urol. 12 (2016) 33–e1.

[12] W.-W. Wang, X.-A. Tu, C.-H. Deng, et al., Long-term sexual activity status and influencing factors in men after surgery for hypospadias, Asian J. Androl. 11 (2009) 417.

[13] F. van den Brink, M. Vollmann, M.A.M. Smeets, et al., Relationships between body image, sexual satisfaction, and relationship quality in heterosexual couples, J. Fam. Psychol. 32 (2018) 466.

[14] V.S. Erikci, A. Sayan, M.O. Ozcan, et al., Glanular approximation procedure in megameatus intact prepuce: a case series, J. Reconstr. Urol. 9 (2019) 95–99.

[15] J. Hardwicke, J.A. Bechar, J. Hodson, et al., Fistula after single-stage primary hypospadias repair - a systematic review of the literature, J. Plast. Reconstr. Aesthet. Surg. 68 (December (12)) (2015) 1647–1655.

[16] S.X. Duan, J. Li, X. Jiang, et al., Diagnosis and treatment of hypospadias with megameatus intact prepuce, Front. Pediatr. 8 (2020) 128.

[17] A. Springer, S. Tekgül, R. Subramaniam, An update of current practice in hypospadias surgery, Eur. Urol. Suppl. 16 (2017) 8–15.

[18] R. Manuele, C. Semni, K. Patil, et al., Foreskin reconstruction at the time of single-stage hypospadias repair: is it a safe procedure? Int. Urol. Nephrol. 51 (2019) 187–191.

[19] M.A. Faasse, D.B. Liu, Early vs. late-presenting urethroplasty complications after hypospadias repair: a retrospective analysis of patient follow-up, J. Pediatr. Urol. 13 (2017), 354-e1.

[20] J.B. Finkelstein, D. Cahill, M.P. Kurtz, et al., The use of telemedicine for the postoperative urological care of children: results of a pilot program, J. Urol. 202 (2019) 159–163.