Received: 20 June 2020 | Revised: 22 July 2020 | Accepted: 27 July 2020

DOI: 10.1111/and.13803

CASE REPORT

First International Journal of Andrology
aNDROLOGIa
WILEY

A closer look at iatrogenic hypospadias

Hüseyin Özbey[1,2] | Değer Devecioğlu[3] | Oleg Staroverov[2]

[1]Department of Pediatric Surgery, Division of Pediatric Urology, Istanbul Medical Faculty, Istanbul University, Istanbul, Turkey

[2]Pediatric Surgery, Division of Pediatric Urology and Andrology, Faculty of Medicine, Sechenov First Moscow State Medical University, Moscow, Russia

[3]Department of Pediatric Surgery, Medi-Life Beylikdüzü Hospital, Istanbul, Turkey

Correspondence
Hüseyin Özbey, Department of Pediatric Surgery, Division of Pediatric Urology (Chairman), Istanbul Medical Faculty, Istanbul University, Millet Caddesi, 34093, Çapa, Istanbul, Turkey.
Emails: hozbey@istanbul.edu.tr; huseyinbey@hotmail.com

Abstract

Almost all surgical repair techniques for hypospadias include dissection of the glans penis, and covering the neo-urethra with the glans tissue circumferentially. Surprisingly, the presence of the "septum glandis" in the ventral midline has been overlooked for decades. A careful examination of six patients with iatrogenic hypospadias (IH) revealed direct indications of the septum glandis. All patients were treated with long-term urethral catheterisation in the paediatric intensive care unit due to neurologic and/or metabolic diseases. The glans was disrupted in all patients due to ventral midline compression of the urethral catheter, which resulted in a tear in the septum glandis. A remnant of the septum glandis was clearly observed in patients with an incomplete tear. Further injuries caused tear in the frenulum and corpus spongiosum, exposed the glanular urethra and made its vertical elliptical shape, the "fossa navicularis", visible. Intact contours of the separated glans wings were observed in all patients. The glans wings merge ventrally in the midline, but are separated by a fine connective tissue (septum glandis) in conjunction with the frenulum, which is involved in the formation of the ventral wall of the glanular urethra. IH provides further insight into the structural anatomy of the normal human glans and glanular urethra.

KEYWORDS

fossa navicularis, frenulum, glans penis, iatrogenic hypospadias, septum glandis

1 | INTRODUCTION

Iatrogenic hypospadias (IH) is a rare condition that occurs in both children and adults as a complication of long-term urethral catheterisation (Andrews, Nauth-Misir, & Shah, 1998; Becker, Witte, Gross, & Netsch, 2018; Mansoor, Ayaz, Rathore, & New, 2016). The downward pressure of the transurethral catheter leads to disruption of the glans penis by harming the "septum glandis". Further injury can lead to various degrees of urethral erosion, namely IH. This type of injury to the ventral male urethra occurs particularly in intensive care units. It cannot be recognised unless it is actively sought, especially in uncircumcised patients.

Almost all hypospadias repair techniques include tubularisation of the urethra over a catheter or stent and approximation of the either limited or extensively dissected glans wings on the ventral midline to enclose the neo-urethra (Hadidi, 2017; Snodgrass & Bush, 2016; Subramaniam, Spinoit, & Hoebeke, 2011). However, careful observation of IH provides direct evidence of the normal anatomy of the glanular urethra and confirms a decades-old misconception in hypospadias surgery.

2 | PATIENTS AND METHODS

Six paediatric cases with IH as a complication of long-term urethral catheterisation were examined. All patients were in intensive care because of neurologic and/or metabolic diseases. The patient characteristics are summarised in Table 1. Detailed photographs of the penises of the patients are taken and evaluated according to the recent studies on normal penis anatomy.

3 | RESULTS

Examinations of the penis of the six patients showed the following findings (Figure 1): (a) In all patients, the glans was disrupted on the ventral midline, but with intact contours; (b) In two patients, the persistent injury was found to be an incomplete glans disruption (patients 1 and 2). The remaining part of the septum glandis was clearly visible in these patients; (c) The details of the internal aspect of the glanular urethra, including the fossa navicularis, were visible in patients with complete disruption of the glans penis (patients 3 and 4) and frenulum, and an injury to the corpus spongiosum (patients 5 and 6). (d) The presence of the foreskin concealed the ventral urethral compression and the injury to the septum glandis (patient 3).

4 | DISCUSSION

The incidence of IH is higher in the elderly patients due to age-related indications of urethral catheterisation such as neurologic and/or musculoskeletal disorders that affect movement and cause urinary retention (Andrews et al., 1998; Becker et al., 2018; Mansoor et al., 2016; Meeks, Erickson, Helfand, & Gonzalez, 2009). In paediatric patients, the foreskin conceals ventral urethral compression and the clinical signs of injury may not be recognised. Nursing staff and intensive care unit doctors should be aware of this condition. A classification of IH has been described recently (Becker et al., 2018). Treatment in the elderly patients is usually in the form of supra-pubic catheterisation, urethral closure and urinary diversion depending on comorbidity (Andrews et al., 1998). In paediatric patients, a urethral reconstruction can be performed if the patient can be discharged from the intensive care unit.

Standard anatomical texts and most studies describe the "glans penis" as the "anterior (expanded) end of the corpus spongiosum" with diagrams showing the urethra that runs through the glans and is therefore surrounded by spongiosal tissue (Cunha & Baskin, 2020; Cunha, Sinclair, Cao, & Baskin, 2020). Dr Friedrich Gustav Jacob Henle, however, first described the gradual termination of the corpus spongiosum on the fossa navicularis (glanular urethra), the ligamentous structure within the glans and its fine fibrous extensions (septum glandis) around the fossa navicularis (Henle, 1877; Özbey, 2019a). The septum glandis divides the glans into outer and inner layers and holds the glanular urethra in the midline through its two main components: the upper median septum and lower median septum. The upper median septum is the extension of the tunica albuginea of the corpus cavernosum penis, which also forms a ligamentous connection (corporo-glans ligament) between the corpus cavernosum and the glans penis. The lower median septum is formed by the extensions of the tunica albuginea of the corpus spongiosum. The lower median septum is connected to the glans between the glans wings and also forms the ventral wall of the glanular and sub-coronal urethra (Özbey, 2019b). Historically, the whole septum glandis was defined as a "urethral ring" and was recently viewed as a "flow control chamber" for urine exiting the urethra (Özbey & Arlı, 2020; von Hayek, 1969). The frenulum is the epidermal extension of the lower median septum, which forms a delta between the glans wings and provides the connection to the foreskin (Henle, 1877; van der Putte, 2007). We recently showed these anatomical details of the glans penis with MRI study (Özbey & Kumbasar, 2017; Figure 2). The development of the glanular urethra is closely related to the development of the foreskin and frenulum in the ventral midline. It is believed that the frustration of the distal-ward growth of the ventral prepuce and frenulum over the urethral meatus results in hypospadias (Liu et al., 2018; van der Putte, 2007). In this study, we show that ventral pressure of the urethral catheter injures the lower median septum and frenulum, resulting in separation of the glans wings from each other, but with intact contours. The glans wings in the normal human penis merge ventrally in the midline, but are separated by a connective tissue (septum glandis) in conjunction with the frenulum. This anatomical feature must be considered in hypospadias surgery, and the glanular urethra should not be covered and compounded circumferentially by the glans wings. The glanular urethra is known to have a wider calibre than the proximal urethra by forming the fossa navicularis. Corpus spongiosum, both in drawings

TABLE 1 The details of the patients

	Age	Diagnosis	Duration of hospitalization with bladder catheter/ year	Type of IH
1	13 months	SMA	22 days/ 1st year	IDGP
2	2 years	Cerebral palsy	30 days/ 1st year	IDGP
3	12 months	RDS, pneumonia, MD	12 months	CDGP
4	17 months	Cerebral palsy, epilepsy	22 days/ 6th year	CDGP
5	6 years	SMA	8 days/ 5th year	CDGPF and CSI
6	5 years	Cerebral palsy, epilepsy	41 days/ 5th year	CDGPF and CSI

Abbreviations: CDGP, complete disruption of the glans penis; CDGPF, complete disruption of the glans penis and frenulum; CSI, corpus spongiosum injury; IDGP, incomplete disruption of the glans penis; MD, metabolic disorder; RDS, respiratory distress syndrome; SMA, spinal muscular atrophy.

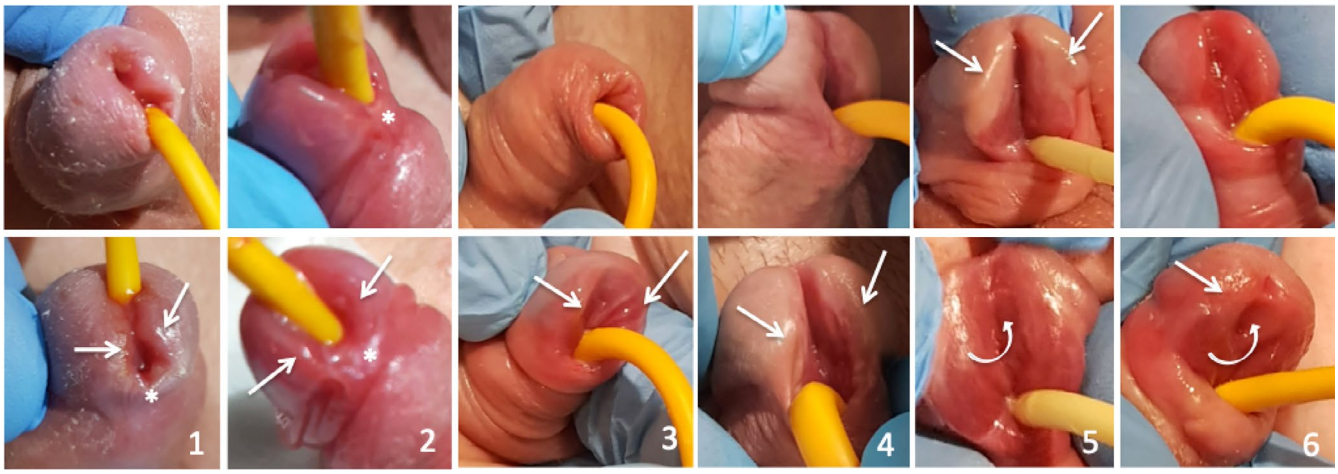

FIGURE 1 Photographs of the patients, 1 to 6. Incomplete disruption of the glans penis (1, 2). Complete disruption of the glans penis (3, 4), and frenulum and injury to the corpus spongiosum (5, 6). Note the remaining part of the septum glandis (*), intact glans wings (straight arrows) and the inner aspect of the glanular urethra showing vertical elliptical shape of the fossa navicularis (curved arrows)

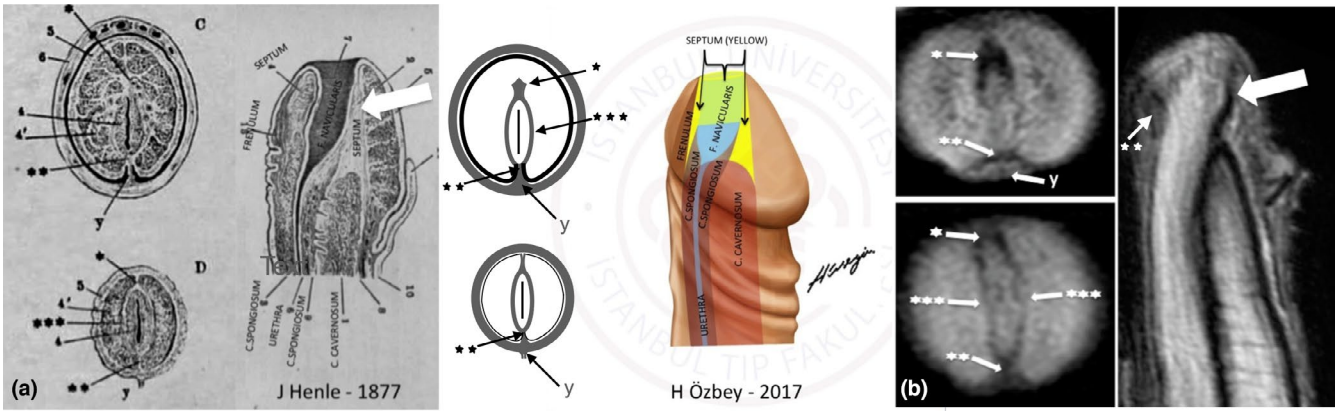

FIGURE 2 (a) Cross sections of the glans penis (C: mid-glanular level, D: sub-meatal level) showing fibrous tissue (septum glandis) surrounding the fossa navicularis (***), its extensions as upper (*) median septum and lower (**) median septum, and the frenulum (y) (Henle, 1877). (b) MR images of the penis show the same anatomical structures and the upper median septum, also known as the "corporo-glans ligament" (large arrow) (Özbey & Kumbasar, 2017)

of Henle and in our MRI study, was detected to gradually terminate at the mid-glanular level where it meets the largest diameter of the fossa navicularis (Özbey & Kumbasar, 2017). At this level, the fossa navicularis forms a vertical elliptical shape that resembles a slit-like passage compressed laterally rather than a round tubular shape. In this study, complete glans disruption (patients 3 and 4) and additional injury of the corpus spongiosum of the proximal urethra (patients 5 and 6) clearly show the elliptical shape of the fossa navicularis, which looks wider than the proximal urethra. These findings are in complete contradiction to the current approaches to urethroplasty, which are almost always carried out in a uniform tubular configuration of the entire urethra (Hadidi, 2017; Özbey, 2020; Snodgrass & Bush, 2016; Subramaniam et al., 2011). We were able to seal the area between the glans wings using the sub-mucosal layer of the ventrally transposed inner prepuce and, like in normal anatomy, form a septum and frenulum (Glanular-Frenular Collar; GFC technique). The GFC technique allows a tension-free tubularisation of the glanular urethra, afforded by limited spongioplasty, which also offers space for the re-formation of the fossa navicularis (Özbey & Etker, 2017).

As a conclusion, the IH provides remarkable information about the anatomical configuration of the glanular urethra, a specific component of the male urethra.

CONFLICT OF INTEREST

The authors have no conflicts of interest to declare. This paper is not published or is under consideration for publication elsewhere.

DATA AVAILABILITY STATEMENT

Data is available at https://doi.org/10.1111/and.13803 to share.

ORCID

Hüseyin Özbey https://orcid.org/0000-0001-6523-1094

REFERENCES

Andrews, H. O., Nauth-Misir, R., & Shah, P. J. (1998). Iatrogenic hypospadias–a preventable injury? *Spinal Cord*, *36*, 177–180. https://doi.org/10.1038/sj.sc.3100508

Becker, B., Witte, M., Gross, A. J., & Netsch, C. (2018). Iatrogenic hypospadias classification: A new way to classify hypospadias caused by long-term catheterization. *International Journal of Urology*, *25*, 980–981. https://doi.org/10.1111/iju.13791

Cunha, G. R., & Baskin, L. (2020). Comments on Professor Hüseyin Özbey's letter. *Differentiation*, *113*, 26. https://doi.org/10.1016/j.diff.2020.03.004

Cunha, G. R., Sinclair, A., Cao, M., & Baskin, L. S. (2020). Development of the human prepuce and its innervation. *Differentiation*, *111*, 22–40. https://doi.org/10.1016/j.diff.2019.10.002

Hadidi, A. T. (2017). History of hypospadias: Lost in translation. *Journal of Pediatric Surgery*, *52*, 211–217. https://doi.org/10.1016/j.jpedsurg.2016.11.004

Henle, J. (1877). *Anatomischer Hand-Atlas zum Gebrauch im Secirsaal*. Band 6. Eingeweide. Braunschweig: Available via University Library Heidelberg, Retrieved from: https://digi.ub.uni-heidelberg.de/diglit/henle1877bd6/0093/image

Liu, X., Liu, G. E., Shen, J., Yue, A., Isaacson, D., Sinclair, A., … Baskin, L. (2018). Human glans and preputial development. *Differentiation*, *103*, 86–99. https://doi.org/10.1016/j.diff.2018.08.002

Mansoor, S. N., Ayaz, S. B., Rathore, F. A., & New, P. (2016). Longitudinal cleavage of the penis in chronic spinal cord injury: Two case reports. *The Journal of Spinal Cord Medicine*, *39*, 366–369. https://doi.org/10.1179/2045772315Y.0000000036

Meeks, J. J., Erickson, B. A., Helfand, B. T., & Gonzalez, C. M. (2009). Reconstruction of urethral erosion in men with a neurogenic bladder. *BJU International*, *103*, 378–381. https://doi.org/10.1111/j.1464-410X.2008.08020.x

Özbey, H. (2019a). The mystery of Jacob Henle's 'septum glandis'. *Journal of Anatomy*, *234*, 728–729. https://doi.org/10.1111/joa.12965

Özbey, H. (2019b). Anatomical relationship between the foreskin and the urethra. *Journal of Pediatric Urology*, *15*, 589–590. https://doi.org/10.1016/j.jpurol.2019.06.027

Özbey, H. (2020). The facts and misconceptions in hypospadias surgery. *Journal of Pediatric Urology*, *16*, 408–409.

Özbey, H., & Arlı, O. T. (2020). "Fossa navicularis" and "septum glandis": A "flow-control chamber" for the male urethra? *Medical Hypotheses*, *140*, 109642. https://doi.org/10.1111/joa.12965

Özbey, H., & Etker, S. (2017). Hypospadias repair with the glanular-frenular collar (GFC) technique. *Journal of Pediatric Urology*, *13*, 34.e1–34.e6. https://doi.org/10.1016/j.jpurol.2016.09.016

Özbey, H., & Kumbasar, A. (2017). Glans wings are separated ventrally by the septum glandis and frenulum penis: MRI documentation and surgical implications. *Turkish Journal of Urology*, *43*, 525–529. https://doi.org/10.5152/tud.2017.00334

Snodgrass, W., & Bush, N. (2016). TIP hypospadias repair: A pediatric urology indicator operation. *Journal of Pediatric Urology*, *12*, 11–18. https://doi.org/10.1016/j.jpurol.2015.08.016

Subramaniam, R., Spinoit, A. F., & Hoebeke, P. (2011). Hypospadias repair: An overview of the actual techniques. *Seminars in Plastic Surgery*, *25*, 206–212. https://doi.org/10.1055/s-0031-1281490

van der Putte, S. C. J. (2007). Hypospadias and associated penile anomalies: A histological study and a reconstruction of the pathogenesis. *Journal of Plastic, Reconstructive and Aesthetic Surgery*, *60*, 48–60. https://doi.org/10.1016/j.bjps.2006.05.020

von Hayek H.. (1969). Der Penis. In K. Conrad, H. Ferner, A. Gisel, W. Krause, S. Wieser, & C. Zaki (Eds.), *Anatomie und embryologie. Handbuch der urologie encyclopedia of urology encyclopedie d'Urologie* (Vol. *1*, p. 372). Berlin, Germany: Springer-Verlag.

How to cite this article: Özbey H, Devecioğlu D, Staroverov O. A closer look at iatrogenic hypospadias. *Andrologia*. 2020;00:e13803. https://doi.org/10.1111/and.13803