

Multiple Circumferential Urethrocutaneous Fistulae as a Rare Complication of Circumcision and Review of Literature

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Circumcision remains the most common surgical procedure performed in boys worldwide. Although circumcision can be associated with numerous major and minor complications, urethrocutaneous fistula is extremely rare and serious. An 18-years-old boy presented with urine passage from 4 fistula orifices. He had been circumcised by nonmedical personnel when he was 2 years old. During the surgery, after degloving the penis, it was observed that the fistulae tracts were combining. There were only 2 fistulae orifices on the urethra. The fistulae were repaired with simple closure. This is the second case reported in the literature describing multiple urethrocutaneous fistulae. UROLOGY 77: 728–729, 2011. © 2011 Elsevier Inc.

Approximately 1 in 3 men are circumcised worldwide, with a higher rate in Islamic and Jewish societies.¹ Although circumcision is the most commonly performed surgical procedure, its complication rate remains high varying, from 1% to 15%.² Fortunately, most of these complications are minor and treated easily without any surgical intervention. However some complications are very serious and generally require surgical treatment. It has been shown that frequencies of complications of circumcision performed by nonmedically trained personnel are higher and that these complications are more serious.³

Urethrocutaneous fistula is a rare complication of circumcision. There are many cases with urethrocutaneous fistula and their surgical treatment techniques reported in the literature.⁴ However multiple circumferential urethrocutaneous fistulae is very rare entity; this is the second case reported.

CASE HISTORY

An 18-year-old boy was admitted to our polyclinic because of multiple urine streams from several openings around the normal meatus. He had been circumcised by nonmedical personnel in his town when he was 2 years old. Physical examination revealed a normally located

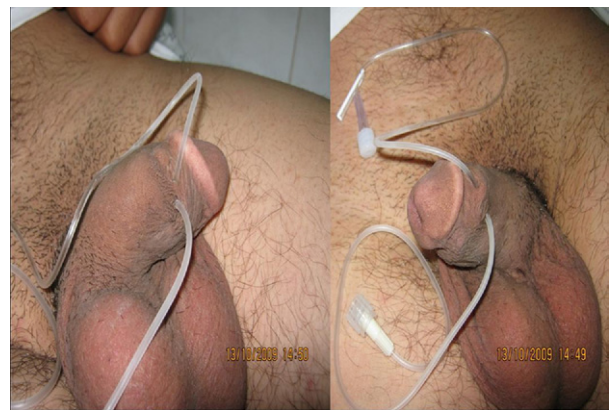


Figure 1. Lateral aspects of the penis, the fistulae are catheterized.

urethral meatus. There were 4 fistulae orifices around corona of penis. On detailed examination, it was observed that there were 2 fistula orifices circumferentially located in both lateral aspects of the penis. Sterile 6F urethral catheters were easily passed through the fistulae Orifices (Fig. 1).

In the operating theater during the procedure, after degloving the penis (Fig. 2), it was observed that the fistulae tracts were combining in each side and creating a large urethral fistula. There were 2 main urethral fistula orifices located on the urethra. The fibrotic tissue was excised, and the fistulae were repaired with simple closure over a 14F urethral catheter. The catheter was removed on the 10th day after surgery. At the last follow-up, the urine stream was in normal calibration, and no fistula was observed (Fig. 3). The patient had no complaints after the surgery.

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Figure 2. Operating theater view of penile shaft after de-gloving procedure, showing fistula orifices.



Figure 3. Postoperative normal urinary stream with no fistula.

COMMENT

Circumcision has become the most commonly performed procedure for medical or religious reasons worldwide, and its benefits are clearly shown. The American Academy of Pediatrics reported that if circumcision is performed before the age of 1 year, the risk of urinary tract infection and penile cancer development decreases.⁵ Despite its benefits, circumcision can be very complicated if performed by inexperienced and nonmedical personnel. The wide spectrum of complications includes bleeding, skin-bridge formation, preputio-glandular fusion, meatal stenosis, chordee, cyst, lymphedema, urinary retention, meatitis, infection, urethral fistula, hypospadias, epispadias, glandular amputation, sepsis, and death.^{3,6,7} Some of these complications are serious and require surgical treatment.

Urethrocutaneous fistula is 1 of these serious complications and is extremely rare. However the rate of urethrocutaneous fistula was reported as 10.4% in a study including severe complications of circumcision.⁶ Although many cases with solitary urethrocutaneous fistula are reported in different series, Agrawal et al published the first reported case of multiple circumferential urethrocutaneous fistulae, located on the dorsolateral aspect of the penis, as a complication of circumcision.⁷ This is the second case reported in the literature involving multiple urethrocutaneous fistulae as a complication of circumcision.

The exact pathologic mechanism of urethrocutaneous fistula is unknown; but nonabsorbable suture materials and very deeply placed ventral sutures or clamping of urethra can lead to this serious complication.

Baskin et al used Mathieu-style skin flap and vascularized penile skin flap to repair the urethrocutaneous fistula. However simple closure of the fistula is another quick and effective method used by other authors.⁶⁻⁸

In conclusion, circumcision is not a procedure to be taken lightly because it can lead to many serious complications. It should be performed by experienced surgeons using absorbable suture materials. In addition, absolute attention should be paid to the procedure, especially during hemostasis and suturing of the ventral aspect of penis to avoid urethral injury.

References

1. WHO/UNAIDS. *Male Circumcision: Global Trends and Determinants of Prevalence, Safety and Acceptability*. Geneva: World Health Organization; 2008.
2. Harrison NW, Eshelman JL, Ngugi PM. Ethical issues in the developing world. *Br J Urol*. 1995;76:93-96.
3. Weiss HA, Larke N, Halperin D, Schenker I. Complications of circumcision in male neonates, infants and children: a systematic review. *BMC Urol*. 2010;16:10:2.
4. Baskin LS, Canning DA, Snyder HM 3rd, Duckett JW Jr. Surgical repair of urethral circumcision injuries. *J Urol*. 1997;158:2269-2271.
5. American Academy of Pediatrics. Tasks force on circumcision. Circumcision policy statement. *Pediatrics*. 1999;103:843-852.
6. Ceylan K, Burhan K, Yilmaz Y, Can S, Kuş A, Mustafa G. Severe complications of circumcision: an analysis of 48 cases. *J Pediatr Urol*. 2007;3:32-35.
7. Agrawal A, Parelkar S, Shah H, Sanghvi B, Joshi M, Mishra P. Multiple circumferential urethrocutaneous fistulae: a rare complication of circumcision. *J Pediatr Urol*. 2009;5:240-242.
8. El-Bahnasawy MS, El-Sherbiny MT. Paediatric penile trauma. *BJU Int*. 2002;90:92-96.