URETHRAL INJURY AS A RARE COMPLICATION OF CIRCUMCISION: A CASE REPORT AND LITERATURE REVIEW

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ABSTRACT

Objective: In this report, we describe the case of urethral injuries after circumcision. Case(s) Presentation: A 5-year-old male came to the urology clinic complaining of urine seeping through the gauze-covered wound. The patient had undergone circumcision. Before the procedure, the patient could generally urinate through the external urethral orifice. His parent realized that the urine was flowing out through the circumcision scar instead of the external orifice. Even though the wound was open and wet, he felt painless when urinating. After cleaning the wound, it appeared that there was a sizeable chromic catgut thread knotted on the right corpus cavernosum. The pendular urethra was cut widely; therefore, the urine came out of this spot. A six French Foley catheter was inserted over the external urethral orifice, the cut urethra towards the bladder. The injured urethra was expected to be attached to simplify the next six-month operation by inserting this urinary catheter. Discussion: Urethral injury post circumcision is rarely reported, but this complication is dangerous due to its morbidity and long-term impact. The paradigm shift from two-stage repair to one-stage repair has developed. Mathieu's modified technique and the Snodgrass technique are recommended. Conclusion: Urethral injury during circumcision can be devastating. The repair technique for urethral trauma depends on the involvement and condition of the surrounding soft tissue, also the size of the tissue damage.

Keywords: Circumcision, complications, fistula, pediatric, urethral injury.

ABSTRAK

Tujuan: Dalam laporan ini, kami menjelaskan kasus cedera uretra pasca sirkumsisi. Presentasi Kasus: Anak laki-laki berusia 5 tahun dibawa orang tuanya ke poliklinik urologi dengan keluhan urin merembes melalui luka yang dilapisi kain kasa, setelah pasien telah menjalani sirkumsisi. Sebelum sirkumsisi, pasien bisa buang air kecil melalui lubang ujung penis. Orang tuanya menyadari bahwa urin mengalir keluar melalui bekas luka sirkumsisi, bukan lubang luar. Meski lukanya terbuka dan basah, pasien merasa tidak nyeri saat buang air kecil. Setelah luka dibersihkan, didapatkan benang chromic catgut yang cukup besar diikat di corpus cavernosum kanan. Uretra pendular terpotong, sehingga keluarlah urin dari tempat ini. Kateter Foley 6 French dipasang dari orifisium uretra eksternum, melalui uretra yang terpotong, menuju kandung kemih. Dengan insersi kateter urin ini, uretra yang ruptur diharapkan dapat berdempetan untuk mempermudah prosedur repair enam bulan ke depan. Diskusi: Cedera uretra pasca sirkumsisi jarang dilaporkan, tetapi komplikasi ini berbahaya karena morbiditas dan dampak jangka panjangnya. Pergeseran paradigma dari perbaikan dua tahap ke perbaikan satu tahap telah berkembang. Teknik modifikasi Mathieu dan teknik Snodgrass direkomendasikan. Simpulan: Cedera uretra selama penyunatan bisa sangat merugikan. Teknik perbaikan trauma uretra bergantung pada keterlibatan dan kondisi jaringan lunak di sekitarnya, juga ukuran kerusakan jaringan.

Kata kunci: Sirkumsisi, komplikasi, fistula, pediatrik, cedera uretra.

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INTRODUCTION

Circumcision is a surgical procedure to remove the foreskin (prepuce) until exposing the glans of the penis. Circumcision is the most commonly performed surgical procedure in the pediatric population. Depending on socio-cultural backgrounds, various sides can perform this procedure, such as skilled healthcare providers, until trained laypersons or rituals. Differences in who

performs circumcision lead to various types and frequencies of complications. Circumcision complication rates vary from 0.2% to 15%.² The most commonly reported complications of circumcision are infection and bleeding at the surgical site.³ Urethral injury is rarely reported, but this type of complication is dangerous due to its morbidity and long-term impact.⁴ Urethral injuries account for 3.4% of overall pediatric genitourinary injuries.³ The prevalence of urethral injury, specifically in circumcision, has not been widely reported.

CASE(S) PRESENTATION

A 5-year-old male came to the urology clinic complaining of urine seeping through the gauze-covered wound. The patient had performed the circumcision three weeks ago before he went to the hospital. This complaint emerged after circumcision in a mass circumcision. It is difficult to trace who and how the operator's competence is because it is done within mass circumcision. Before the patient is circumcised, the patient can urinate normally through the meatus urethrae external. The patient is a student, and he does not have a history of particular disorders or diseases. There is no specific illness in his family illness history. Several days after the circumcision, the patient and his family realized that each time the patient's urine did not come out

through the urethral meatus but the urine through the circumcision scar. We can see the urine from the gauze covering the wound, which is always wet and smelled of urine. The patient does not experience pain when urinating.

The patient had twice follow-up treatments with the local midwife. The patient only received regular wound care, namely dressing gauze, antibiotics, and pain reliever. After three weeks of home treatment, urine never came out through the meatus urethrae external. Finally, the patient went to the secondary hospital.

On physical examination, it appears that the wound is open and wet, urine fluid comes out of the injury (Figure 1A). No urine passes through the meatus urethrae external. After cleaning the wound, we found a sizeable chromic catgut thread knotted on the right corpus cavernosum and penetrate the urethral tissue. The incision is cut through the pendular urethra widely so that urine comes out of the damaged urethral tissue (Figure 1B). The glans penis is intact (Figure 1C). We suspect that the operator accidentally incised the corpus cavernosum, whereas this area may cause much bleeding. This incident may be influenced by several factors, such as the lack of operator competence and the absence of proportional bleeding control instruments, so the operator decides to do ligation using a large size of chromic catgut. The urethra was partially ruptured due to inappropriate ligature.

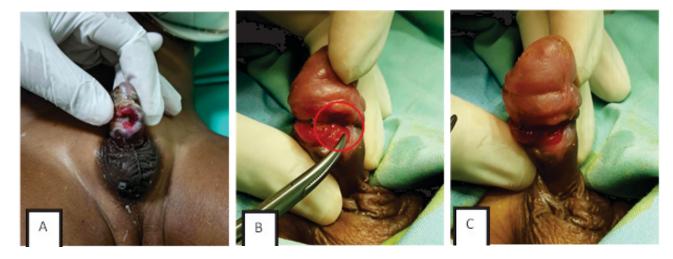


Figure 1. (A) The clinical appearance on hospital admission. (B) The red circle shows the unconnected urethra (C) The glans penis is still intact.



Figure 2. The Urologist inserted a 16F urine catheter to connect the two unconnected urethrae.

The patient is planned for surgical repair under general anesthesia. The Urologist has six years of experience in the urology field. After cleaning the circumcision scar, a 6 Fr catheter is inserted through the external urethral meatus and then into the cut urethra until it continues to enter the bladder (Figure 2). During post-operative treatments, the patient gets analgesic therapy if needed, and the patient is routinely coming to the urology clinic for wound care and dressing for urine catheter. The patient and his parents are also educated to maintain hygiene in the penis area. At a 1-month follow-up visit, the wound around the urethra has dried and has become a urethrocutaneous fistula (Figure 3). The diameter of the fistula is about 5 mm. The patient has a urethrocutaneous fistula closing procedure under general anesthesia. At the post-operative visit, the patient said there was no seeping urine with minimal pain. The patient's parents are also satisfied with the function and cosmetics of the patient's penis. Patients will continue to be evaluated periodically in the next six months

DISCUSSION

Severe complications after circumcision, such as urethral trauma, are reported to be very rare. Reported severe complications rates of circumcision vary from 0-2%. Some of the proposed risk factors associated with circumcision severe complications are older patient age, lack of operator experience, procedures performed by untrained providers, incomplete detachment around the frenulum balano-preputial adhesion, and the factor of the instrument

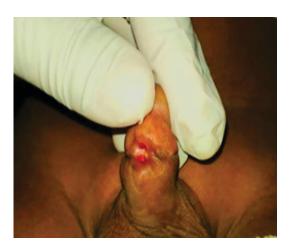


Figure 3. The condition of urethrocutaneous fistula on a one-month follow-up visit,

used. Some types of clamp instruments increase the risk of injuring the penile tissue because the provider cannot directly visualize the penile tissue before cutting the prepuce.⁴ The previous study stated that the complication rate in circumcision performed by non-health professionals or untrained providers was around 80%.⁷

Depending on the injury level, urethral injury after circumcision may not be identified initially and developed in the following days. Accepted mechanisms of the urethral complication in circumcision are including (1) excessive prepuce traction, which increases the risk of cutting the ventral urethra during prepuce cutting; (2) uncontrolled sutures which are performed to control bleeding in the frenulum area; (3) poor hygiene control after circumcision, leading to severe fasciitis. 9

Based on the literature review (Table 1), penile trauma can be diagnosed by anamnesis and physical examination on the damaged tissue (commonly in the form of glans amputation or even urethral trauma). A layperson who does not have a medical background, even physicians and urologists, may experience complications when performing a circumcision procedure. Centikaya et al., Faydaci et al., and Raisin et al. found 11 cases of penile trauma in children performed by a layperson. In comparison, Khaireddine et al. and Manentsa et al. found 6 cases of trauma due to circumcision performed by operators with medical backgrounds. Patients' prognostic depends on the severity of the trauma, the early treatment, and postoperation treatments. Post-reconstruction surgery

Table 1. Summary of complication following a circumcision procedure.

Paper reference number and year	Total of patients	Age of patient	Symptoms and clinical presentation	Circumcision operator	Method of diagnosis	Treatment	Follow up time and prognostic information
Centikaya et al. ¹⁶ 1992	2	2,5 y.o	Both of them have no ejaculation and erection; the glans penis and corpus cavernosum were invisible, external urethral meatus at the	Itinerant circumcision	Clinical Presentation	Reconstructed the penis with a suprapubic graft and released the corpus cavernosum at their 19 years old	Six months; they can have sexual intercourse
			mons pubis; defects of the glands and corpus, iatrogenic hypospadias.				
Faydaci et al. ¹⁴ 2010	1	7 y.o	The patient has amputation of the glans penis in the one-third distal part. The prepuce was excised completely (Clamp technique)	No medical background	Clinical presentation	Primary reattachment technique and hyperbaric oxygen therapy	After two months, no meatal stenosis, normal voiding, good cosmetic
Khaireddine et al. ¹⁷ 2014	2	3-5 y.o	The complete amputation of gland penile; Partially amputation of gland penile. In both cases, the prepuce is pulled and clamped just distal to the glans of the penis. The operator makes cuts in the area between the clamp and the gland	General practitioner and Urologist	Clinical presentation	Non-microsurgical reattachment of distal penile glans	After two years, no stricture, no fistula, excellent cosmetic and routine function

Table 1. (continues)

Paper reference number and year	Total of patients	Age of patient	Symptoms and clinical presentation	Circumcision operator	Method of diagnosis	Treatment	Follow up time and prognostic information
Manentsa et al. ¹⁰ 2019	3	10 – 15 y.o	1 The complete glans amputation; 2 with the partial glans amputation (Forcepsguided technique)	Medical officer, nurses, and clinical associates	Clinical presentation	Repair and reconstruction penis	A fistula and had to undergo next reconstructive surgery; A urethra cutaneous fistula; Healed without sequelae
Raisin et al. ² 2020	8	7-9 days	amputations of distal glans; 4 amputations distal glans + urethra; 1 complete glandular amputation; and 1 distal glans + meatal amputation	Laypeople (ritual circumcision)	Clinical Presentation	The Urologist performed traditional reconstructive plastic surgery + intraoperative debridement in nonviable tissue. Careful reapproximating of the edges of the trauma tissue was performed.	Two patient hypospadias
Soltani et al. ¹⁸ ., 2020	1	4 y.o	The complete glans amputation (the Mogen clamp technique)	General practitioner in an outpatient office	Clinical presentation, Ultrasound, urinary flowmetry	Penile shaft replantation and pentoxifylline	Twenty-one days after the operation, the Urologist removed the Foley catheter, and the appearance penis was good. Three months later, regular voiding stream, no stenosis, satisfactory cosmetic

complications include hypospadias, urethra cutaneous fistula, and cosmetic problems. ^{2,10}

Because urethral injury without glans involvement is rare in circumcision, no single method is recommended to manage this type of

urethral injury. Some authors report secondary healing methods, with delayed repair when needed. In certain conditions, a primary anastomosis is recommended.⁴ As urethral reconstruction techniques have developed, a paradigm shift from

two-stage repair to one-stage repair has developed. This method can use a graft free of the buccal mucosa and skin or a combined approach.¹¹ The technique of repairing a urethral injury depends on the degree of damage, the location, the structure of the penis, and the condition of the surrounding soft tissue.¹² It depends on the patient's status, demanding immediate improvement; Mathieu's modified technique and the Snodgrass technique are recommended.¹³

As in our case, primary urethral anastomosis with a Foley catheter was recommended for patients with post-circumcision urethral injury. This procedure aims to maintain the meatal position and continuity of the urethral tissue. The placement of a foley catheter will reduce the need for further surgical intervention and prevent secondary hypospadias. Urethral anastomosis with this technique can still cause complications such as urethral-cutaneous fistula, urethral stricture, or chordee - if there is a shortening of the urethral tissue.¹⁴

Eventually, there is a moral obligation to reduce the risk of harm associated with any surgical procedure. In a circumcision, it is necessary to ensure that the provider who performs the circumcision has adequate training (doctor, nurse, or traditional-religious provider) and has measured competence. Some of the other approaches needed to minimize the risk of complications after circumcision are optimal infection control and pain management during and after the procedure. ¹⁵

CONCLUSION

Circumcision is a safe surgical procedure performed by a competent operator. Severe complications, such as urethral injury, are more likely to occur in conditions where inexperienced providers perform them. Although they are infrequent, urethral injury during circumcision can be devastating. Surgical repair of urethral injury depends on the involvement and condition of the surrounding soft tissue. In our case, it provides good functional and cosmetic outcomes.

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