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<http://www.pediatricurologycasereports.com>**A rare complication following circumcision, severe urethral fistula located on the distal penile shaft and the coronal margin: A case report****Ahmet Atıcı, Mehmet Emin Çelikkaya, Bülent Akçora***Department of Pediatric Surgery, Mustafa Kemal University, School of Medicine, Antakya, Hatay, Turkey***ABSTRACT**

Fistula following circumcision is a serious problem when applied by especially untrained professionals. We present a two-year-old male patient with severe penile and urethral trauma developing fistula after circumcision. Physical examination revealed that the patient had severe tissue defect starting from the glans penis inferior and reaching up to proximal penile level and including the ventral lateral sides of the urethra, and wide urethral fistula. Circumcision is considered as a benign intervention but its rare complication as fistula is sometimes severe and the treatment delicate.

Key Words: Circumcision; complications; fistula; child.

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Introduction

Urethral injuries occur in 3.4% of genitourinary injuries in children [1]. Circumcision is the surgical removal of the skin called preputium, which covers the tip of the penis, to reveal glans penis. Circumcision is the most frequent surgical operation of childhood [2]. Although benefits of circumcision are still controversial, it is accepted due to its approved benefits such as reducing the risk of HIV transmission, facilitating urination, reducing the risk of cancer, providing better cosmetic image, and

increasing penile hygiene [3]. Although there are numerous reports of circumcision complications, the most frequently known and reported complication is hemorrhage and wound site infection [4, 5]. Urethral fistula is not a very common complication [6, 7]. In this paper, we present a two-year-old male patient with severe penile and urethral trauma with severe urethral fistula after circumcision.

Case report

Two-year-old male patient presented to the emergency service of our hospital 5 days after his circumcision in Syria, with complaints of wound infection and discharge in his penis and urinating downwards. Physical examination revealed that the patient had severe tissue defect starting from the glans penis inferior and reaching up to proximal penile level and

including the ventral lateral sides of the urethra, and wide urethral fistula [Fig. 1].



Fig. 1. Severe tissue defect starting from the glans penis inferior and reaching up to proximal penile level and including the ventral lateral sides of the urethra.

The patient was peeing from the neomea at the newly formed wide urethral defect starting from the bottom of the corona level. Patient was prescribed with antibiotic (ampicillin sulbactam) and nonsteroidal anti-inflammatory drug for one week. Definitive operation was scheduled 6 months later.

Discussion

Although it is considered as an easy surgical operation, circumcision has a variety of complications. In numerous prospective studies, the reported rate of complications after circumcisions is between 0- 30% [8]. The most frequently recognized complication is hemorrhage, whereas there are publications which even present cases of death following the circumcision-related complication and sex change [2]. Many complications such as

wound infection, sepsis, painful erection, concealed penis, incomplete circumcision were reported [4]. Severe complications such as urethral trauma, penile trauma and glans amputation after circumcision are reported very rarely [2,9]. The rate of severe complications varies between 0-2% in different studies [8]. Our patient's injury was diagnosed as severe urethral and penile injury [Fig. 1]. Among these complications, one of the most troublesome is urethral fistula [10]. According to a recent study, the diameter of urethral fistula was between 1.5-12 mm, and recurrence rate was higher in larger fistulas [11]. Our patient's urethral trauma was too large. Urethral injury after circumcision can vary from country to country in various studies. For example, in our country where circumcision rate is almost 100%, number of reported urethral injuries is close to zero [4], whereas it is very high in sub-Saharan Africa [11]. The reason behind this high rate in Africa is the failure to provide sterile conditions and the fact that people who are not doctors perform the circumcision [11]. In one study, complication rate in circumcisions performed outside the hospital by someone who is not a doctor is reported to be 80% [12]. In our case, severe complication that our patient experienced was attributed to unsterile conditions and insufficient equipment for circumcision operation, given the fact that there is an ongoing war in Syria. Urethral injury after circumcision may not be recognized in the early stages, and can develop in the following days in perineal, scrotum, penile, corona, or glans penis depending on the injured level [13]. Urethral trauma is observed more frequently after newborn circumcisions, as the urethra in the coronal level is superficial and closely related to frenular veins in the newborn [10]. While urethral fistula is

generally located at a single area such as corona or penile, the trauma was started from beneath of glans penis and reached up to proximal penile level, in present case [Fig. 1]. Excessive traction of preputium during circumcision can cause ventral urethra to be cut together with the prepuce [4,7,10]. Anamnesis of the patient showed that he had severe swelling and wounds on his penis on his first day and that he urinated from downwards. The etiology of the developing complication was attributed to the failure to notice the urethral trauma that occurred preoperatively in the early stage, and the additional infection. In the etiology, uncontrolled sutures performed to control the hemorrhage that occurs in the frenular region can cause urethral traumas. Severe fasciitis that develop due to bad hygiene after circumcision is accepted as another etiological cause [10]. In surgical treatment of urethral injuries, repair, which is a single-stage operation that provides good cosmetic results and sufficient penis size and function, is considered as the golden standard in urethral injuries [11]. Repairing the urethral injury, which is noticed perioperatively, in the same session gives very good results [11]. However, treating traumas that are noticed later are harder to treat. In order to obtain better results in urethral traumas noticed post-operatively, postpone of the operation for a minimum of 6-months is recommended for the inflammation to cease and completely be cured [11]. Repair technique depends on the localization of the injury, size of the urethral damage, state of the surrounding soft tissue, preputium status and penis structure. Mostly, multilayered, tension-free closure is required [14]. For these patients, depending on the status of the patient, primary repair, modified-Mathieu's technique and Snodgrass repair are recommended. In some studies, cases where

urethral mobilization is followed by end to end anastomosis, were reported [15]. Since our patient did not have sufficient dartos layer, preputial tissue, subcutaneous tissue and skin, and also due to the fact that he urinated from the newly formed proximal neomea, it was planned that the patient will be handled in the definitive operation as if he was a case of circumcised proximal hypospadias.

Conclusion

Surgical circumcision is a very safe operation at experienced hands. However, in inexperienced hands and under circumstances where the hygiene conditions are not sufficient, severe complications such as urethral and penile trauma can be seen, although rarely. Management of wide urethral fistulas is hard and recurrence rate is high. Therefore, we think that preventing the development of such complications after circumcision is as important as treating them.

Compliance with ethical statements

Conflicts of Interest: None.

Financial disclosure: None.

Consent: Written informed consent was obtained from the parent of the patient.

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