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Near-total penile amputation secondary to hair Torniquet syndrome – Case of a staged repair with a satisfactory outcome

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ABSTRACT

Penile Hair Tourniquet Syndrome, a subtype of Penile Tourniquet Syndrome, is a rare condition and a pediatric surgical emergency seen in infants whereby a constricting hair coil strangulates the penis most often at the coronal sulcus, leading to edema, ischemia and necrosis, If untreated, it can potentially lead to development of a urethrocutaneous fistula or even urethral transection and penile amputation. Therefore, a well-timed intervention can prevent complications and lifelong unhappiness. Herein, we present a case of an 11-month-old boy who presented with near total amputation of the glans and underwent two-staged repair with a satisfactory outcome.

1. Introduction

Penile Hair Tourniquet Syndrome (PHTS) is a rare condition that is typically seen in infants whose mothers are experiencing postpartum hair loss, known as telogen effluvium.[1,2] Several case reports have been published on this condition; however, very few have been reported from Pakistan. The most serious complications of PHTS include ischemia, necrosis, urethrocutaneous fistulas, gangrene, and even auto-amputation.[3]

In this report, we present a case of an 11-month-old boy who presented with near-total amputation of the glans and underwent a two-staged repair with a satisfactory outcome.

2. Case report

An 11-month-old circumcised boy presented with complaints of an abnormal appearing glans noticed by his father the day before. He was passing urine from an opening on the ventral surface of the penis, and the glans was hanging by a narrow pedicle.

On examination, the glans was pale-looking and attached at the partly amputated left corpus cavernosum. A constricting hair was found at the coronal sulcus (Fig. 1A). A complete transection of the right corpus cavernosum and corpus spongiosum along with the urethra was noted (Fig. 1B).

The child had a concomitant chest infection. The constricting hair was removed under sedation and penile block (Fig. 1C). A size 6Fr feeding tube was passed from meatus crossing the distal to proximal urethra. Tacking sutures were applied in four quadrants with 5/0 vicryl to prevent further damage secondary to torsion on a narrow pedicle. Subsequently he was treated for upper respiratory tract for 2 weeks.

Definitive surgery was planned two weeks later. Under general anesthesia, a penile tourniquet was applied at the base of the penis to control hemorrhage. Tacking sutures were removed. The penis was circumferentially incised over the circumcision scar and degloved to the mid-shaft. The urethral edges were mobilized up to 3 mm, and circumferential urethroplasty was performed over a 6Fr silastic Foley catheter with 6/0 PDS. A dartos flap from the penile shaft was used to cover the urethral repair ventrally. The glans and opposing corporal surface were de-epithelialized and sutured together with 6/0 PDS. The degloved penile skin was pulled forward and sutured to the subcoronal margin circumferentially. A thick Duoderm dressing was applied.

The postoperative period was uneventful. The dressing was removed on day 4 and the catheter was removed on day 12. The patient was recovering well at 2-weeks postop (Fig. 1D). Postoperative follow up at 6 months showed a satisfactory cosmetic and functional outcome (Fig. 1E).

3. Discussion

Two of the most common causes of pediatric penile trauma are circumcision and penile strangulation, also known as penile tourniquet syndrome. Hair coil strangulation is the most common cause of this

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Fig. 1. A, Near total glans penis amputation with a visible hair tourniquet. **B**, Penis after removing the hair tourniquet. **C**, Ventral view of the penis showing a completely transected urethra. **D**, Penis at the 2 weeks postoperative period. **E,** Voiding of urine at 6-month follow-up.

Table 1
Grading of injury in penile hair tourniquet syndrome (as described by Bashir and El-Barbary).

Grade	Description of the injury
Grade 0	Constriction of skin, without urethral injury
Grade I	Partial division of the corpus spongiosum and occurrence of a urethrocutaneous fistula
Grade II	Complete division of the corpus spongiosum and constriction of the corpus cavernosum
Grade III	Gangrene, necrosis, and complete amputation of the glans penis

syndrome, accounting for 79% of all cases.[4] PHTS is often seen in circumcised boys due to the exposure of the coronal sulcus and it is characterized by the strangulation of the glans penis by a constricting hair coil. Initially, lymphatic and venous backflow are affected, leading to edema and swelling of the distal penis, which can mimic balanitis. This is followed by varying degrees of injury, which can result in the development of a urethrocutaneous fistula (UF), urethral transection, and partial or complete amputation of the glans. Division of dorsal nerves may also alter sensations of the glans, which can affect sexual experiences later in life.[5] Bashir and El-Barbery have classified the degrees of urethral injury in PHTS into four grades, as shown in Table 1.[6] In our case, the injury falls between grade 2 and 3.

Although around 100 cases of PHTS have been reported in the literature,[7] the condition is still rare enough to be recognized promptly.[4,8] Vague presenting symptoms, neo-epithelialization over the constricted region and clinical appearance resembling balanitis can deceive an experienced clinician.[9] Therefore, a thorough examination and high index of suspicion is crucial in making a diagnosis. Differential diagnoses to consider include insect bites, infection, trauma, dermatitis, or congenital constriction bands. If any visible depression (i.e., hourglass deformity) or sharp demarcations are seen, probing should be performed at the lateral aspects of the penis to isolate constricting hair strands which may not be apparent at first sight. Urgent referral to specialists for examination under anesthesia is warranted if removal is not easily achieved. Well-timed intervention can prevent complications and lifelong unhappiness.[10]

Surgical procedures for PHTS depend on the grade of injury.[6] Grade 0 injury warrants removal of constricting material only, whereas Grades I and II require formal surgical repair. In Grade III injury, replantation of the glans, as reported by Facio et al. may be possible.[11] This can be done as a single-stage repair or delayed repair. Badawy et al. reported 4 complications in 25 patients (16%) who underwent single-stage repair (2 strictures and 2 fistulas), which were successfully repaired without recurrence.[4] Harouchi et al. performed a single-stage repair in most of their 38 cases, except for the severe ones where the glans was hanging to the penis by a very slim pedicle, in which case the glans was re-anastomosed to the corpus initially and then a delayed urethroplasty was performed.[12]

Similarly, in our case, we performed a rescue four quadrant stitching and stenting of the urethra to prevent twisting of the pedicle with complete loss of glans. Definitive surgery was planned after proper optimization of the patient. In our experience, tacking sutures are essential to prevent further injury due to twisting or torsion while the patient is being optimized for definitive repair. Moreover, in acute cases, these tacking sutures can play a useful role in buying time to resolve edema.

4. Conclusion

PHTS is a surgical emergency that requires immediate intervention to prevent potential complications. Early recognition and urgent urologic consultation are crucial. Glans tacking sutures are a useful option to gain time for definitive repair.

Conflict of interest statement

All authors declare that they have no confict of interest. All authors have no funding sources to declare.

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