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Coronal urethrocutaneous fistula in an uncircumcised child with penile hair tourniquet syndrome: A case report

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ABSTRACT

Hair tourniquet syndrome is characterized by circumferential strangulation of an appendages or genitalia by human hairs or fibres. Here we are reporting a rare case and its successful management of coronal urethrocutaneous fistula developed due to penile hair tourniquet syndrome (PHTS) in a 5 years old child.

Key Words: Penile hair tourniquet syndrome; urethrocutaneous fistula; fistula repair; child.

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Introduction

This pathological condition first described in 1832 and in 1971 termed as hair tourniquet syndrome by Quinn [1]. It occurs when a strand of hair, or occasionally a piece of thread or fibre encircles an appendage, causing partial or total obstruction to circulation. The condition is very rare that occurs mostly in children. Penile hair tourniquet syndrome (PHTS) causes progressive strangulation and may result in various complications from urethra-cutaneous fistula, complete urethral transaction and penile gangrene to glans amputation.

Case report

A 5 years old boy, presented to us with complaints of pain and swelling over the penis, and difficulty in urination for the last one month. The baby was uncircumcised and belongs from Hindu family. On careful examination of child penis after retracting the prepuce, a band of hair tourniquet was seen at coronal sulcus causing coronal fistula and circum-coronal band of inflammation and ulceration [Fig. 1 A and B]. The glans, rest of the penis, bilateral testis and scrotum were normal. On further examination, the child was average built for his age. Abdominal examination was normal and the bladder not palpable. Despite details history we could not found the cause of this problem (including the child abuse, sexual assault or any foul play). Under sedation the hair tourniquet was

removed using the blunt probe cutting technique and child was discharged on analgesics and antibiotics [Fig. 1C]. On further follow up of child the ulceration and inflammation of coronal sulcus were improved, but the coronal fistula was not healed till four months [Fig. 1D and E] so surgical repair of coronal fistula performed using the dartos flap as a second layer cover [Fig. 1E]. Postoperatively, the patient did well up to one year of follow up.

Discussion

PHTS is a very rare clinical situation. In this situation, fibers of hair tied around the penis. The cause may be accidental, incidental, intentional, or indeterminate. Circumcision as a predisposing factor as a hair is more easily entangled around a circumcised penis than around a glans covered by an intact prepuce [2]. This condition generally occurs in children. In our case the child was five years old and the symptoms was for around one

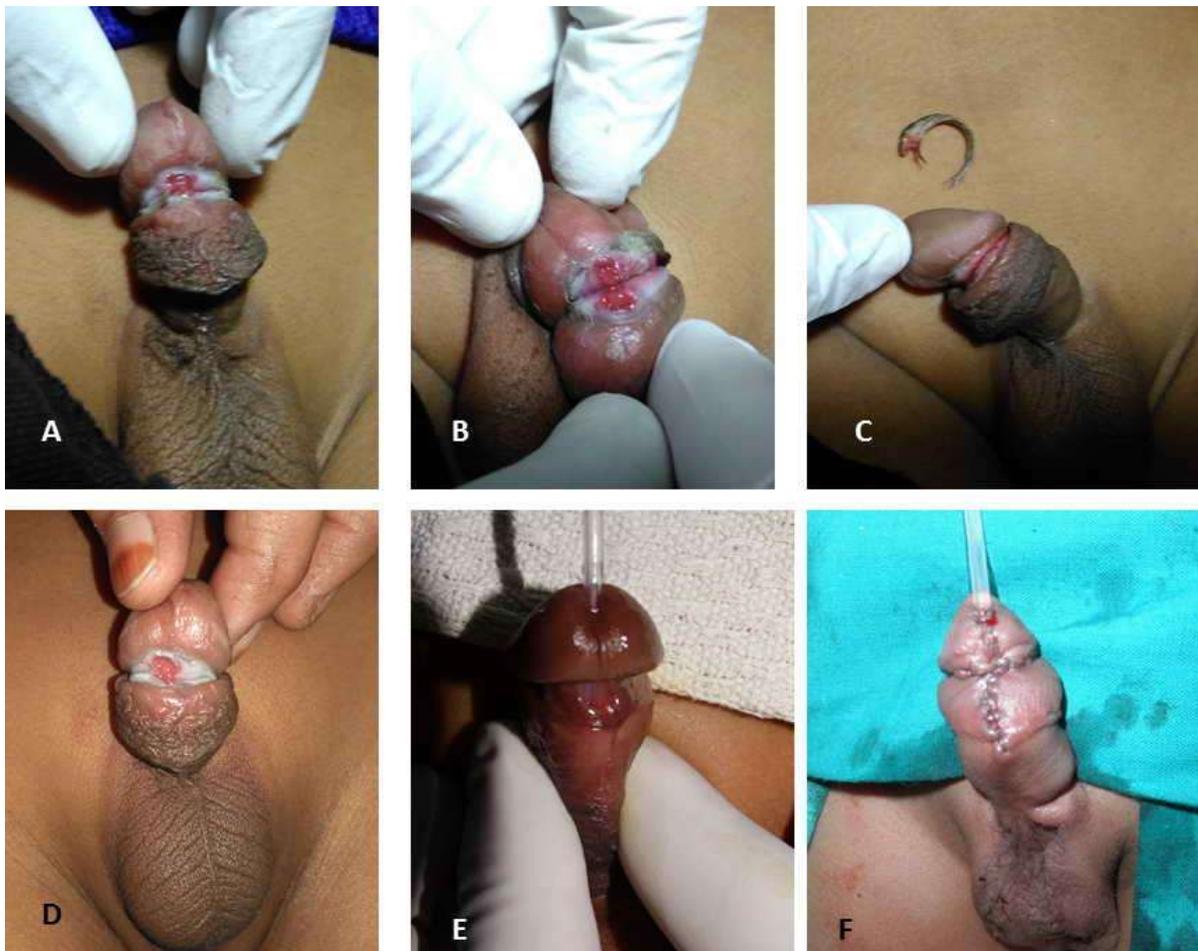


Fig. 1. A and B) Penile hair tourniquet at corona with urethrocutaneous fistula, prepuce is retracted. **C)** Coronal injury after removal of hair band. **D)** Healed injury after one month of hair tourniquet, only coronal urethrocutaneous fistula is present. **E and F)** Preoperative and post-operative look of penis.

month. Though the condition was reported almost exclusively in circumcised boys but our case was exception to this rule as our case was not circumcised and hair tourniquet was only visible on retracting the prepuce (Fig. 1 A and B) [2]. In our cases, we could not find the cause after detail history and inquiry to parents.

The presentation of this entity either may be in acute over day or insidiously over months. In the latter instances, as in our case the fiber can cut-through and become buried under the skin, which may re-epithelialize obscuring the offending hair altogether. The ring of hair has a high tensile strength and stretches when wet and contracts while drying, it applies a tightening constriction of the structure and leads to injury. The pathology of injury are usually the constricting ring on the organ cause vascular and lymphatic obstruction leads to tissue edema causing necrosis, ulceration and tissue loss. Bashir et al [3] in 1980 have categorized severity of penile strangulation as follows: Grade 0: Constriction of skin without urethral injury, Grade 1: Partial division of corpus spongiosum with urethro-cutaneous fistula, Grade 2: Complete division of corpus spongiosum and constriction of corpus cavernosum, Grade 3: Gangrene, necrosis and complete amputation of glans. In our case, the injury falls into the grade 1 of above classification.

According to a recent study, authors reviewed a total of 210 reported cases of hair tourniquet syndrome and found that penile involvement occurred in 44.2%, and in another study hair remains the most common causative agent in 79% cases [1,4]. Early recognition of this condition is important and requires a high index of suspicion when no other causes found in an irritable child with urinary discomfort. The causative agent (hair) should be removed promptly to avoid further damage. Techniques

described are the unwrapping technique, the blunt probe cutting technique and the incisional approach as in our case we used blunt probe cutting technique [5,6]. Once the constricting band is removed further repair should be done after improvement of edema and inflammations. Depending on the severity of the injury, urethrocutaneous fistula repair, end-to-end urethroplasty or urethral advancement and glanuloplasty incorporated procedure may be used [5]. In our case as there was only a coronal urethrocutaneous fistula, so we repaired the fistula interposing the dartos cover.

Conclusion

PHTS, especially in a child, is a rare and serious clinical condition. High index of suspicion is needed to identify the condition. Early diagnosis and appropriate treatment can prevent the loss of glans and other complications. Child abuse should have considered carefully in suspected cases.

Compliance with ethical statements

Conflicts of Interest: None.

Financial disclosure: None.

Consent: All photos were taken with parental consent.

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