

An Unusual Cause of Urethrocutaneous Fistula in a 10-year-old Boy

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Introduction

10 year old boy, previously circumcised, was noticed by the attendants to have multiple strands of hair tied around the penile shaft, moreover the father noticed an abnormal spraying during urination. On examination; there was a band of hair around the coronal sulcus, along with penile engorgement and swelling, there was an abnormal opening ectopically located on the ventral aspect of the penis, proximal to the tip of the glans (Figure 1). Patient was taken for surgery (Figure 2), and was discharged home the next day.



Figure 1: (A) Hair band around the coronal sulcus complicated by an abnormal opening at ventral aspect of the penis (B)



Figure 2: Intraoperative repair of the defect

Questions:

- 1-What is the likely diagnosis of this patient ?
- 2-What are the complications associated with this syndrome ?
- 3-How would you manage this case ?

Answers:

- 1-Penile tourniquet syndrome
- 2- Urethrocutaneous fistula as in this case , complete urethral transection, penile gangrene and amputation
- 3-This patient should be managed surgically by removing the hair band and by repairing the urethrocutaneous fistula using a well vascularized penile dartos-based flap

Discussion:

Tissue strangulation by a thread was first introduced by Guillimeau in 1612 (1) . Hair tourniquet syndrome (HTS) occurs when strands of hair entangle around fingers, toes and rarely the penis. In penile tourniquet syndrome (PTS) a hair coil is found to be wrapped around the sulcus coronarius of the penis. This is usually attributed to the fact that mother's hair tends to fall (telogen effluvium) on the baby's genitalia while changing diapers. It usually happens in circumcised babies between the age of 0 to 6 years (2), however ,our patient was diagnosed at the age of 10 years and there are few reported cases at this age . Complications associated with PTS includes ; urethrocutaneous fistula, complete urethral transection, penile gangrene and amputation. The pathophysiology behind PTS is attributed to the lymphatic obstruction from tourniquet effect of the hair that leads to tissue edema which causes venous outflow obstruction and eventually impairs the arterial perfusion resulting in necrosis and tissue loss . Anatomical properties of the penile shaft and its

association with strangulation was described by Bashir et al (3), he categorized penile strangulation as:

Grade 0: Constriction of skin without urethral injury

Grade 1: Partial division of corpus spongiosum with urethrocutaneous fistula

Grade 2: Complete division of corpus spongiosum and constriction of corpus cavernosum

Grade 3: Gangrene, necrosis and complete amputation of glans.

PTS is misdiagnosed in most of the time and this happens because most of the physicians are not alert of it. Diagnosis is made clinically; patients usually present with unexplained penile swelling or pain, purulent discharge, urine leakage from coronal sulcus or a gangrenous glans. Clinicians may face difficulty in identifying the hair band as it is usually covered by the swollen skin. Complications such urethrocutaneous fistula and penile gangrene can be avoided by early removal of the hair band; this can be done under local or general anesthesia. In children who present with urethral fistula, as in our case, direct closure of the defect by a well vascularized penile dartos-based flap is usually the preferred procedure (4). In severe cases where the glans is separated from the penis , the repair can be done in two stages ; glans anastomosis to the corpus first, followed by urethro- plasty at a later stage (5). Some studies advocated the use of topical agents that contains thioglycolate, calcium hydroxide or sodium hydroxide to dissolve the hair atraumatically .

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