Penile hair tourniquet resulting in hypospadias failure

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ABSTRACT

Penile hair tourniquet (PHT) is a painless form of penile ischemia, typically seen in toddlers with long-haired mothers, caused by entanglement of hair on the balano-prepuce sulcus, normally associated with circumcision. Its association with hypospadias has been reported only once. A school-aged boy admitted for surgery to treat hypospadias failure was incidentally detected to have PHT and severe hourglass deformity of the penis. Urethral anastomosis and glanuloplasty were done after removal of the constricting ring, without complications. Normal erections were reported during follow up. Treatment may involve urethral reconstruction and penile reimplantation in extreme cases.

Key words: Hair, hypospadias, ischemia, penis, reconstruction, tourniquet

INTRODUCTION

Penile hair tourniquet (PHT) has been described as a rare cause of penile ischemia, mainly in circumcised toddlers. Most cases have been attributed to the inadvertent entrapment of hair strands from long-haired mothers on the babies’ penises. The problem has frequently been diagnosed late despite frequent inspections of the baby’s genitalia while changing diapers.

We report a case of PHT associated to hypospadias in a school-aged boy, and discuss its implications on the treatment of the hypospadias and long-term consequences of the ischemic episode.

CASE REPORT

A 10-year-old boy was admitted for surgery to treat hypospadias failure after multiple previous surgeries. The patient originally presented with penisocrotal hypospadias, was treated by orthoplasty and an onlay (Duckett) procedure. A series of surgeries followed in order to treat meatal stenosis and recurring urethral fistulae. The present surgery was scheduled to treat a large recurrent distal urethral fistula and glans disfiguration. Pre-operative testosterone was prescribed (3 monthly injections, the last one to be administered 15 days before surgery).

At admission, the patient denied new complaints and reported the usage of three injections of depot testosterone as prescribed. Physical examination showed a small and disfigured glans and a tight hair tourniquet lodged in the balanoprepuce sulcus, causing serious narrowing of the corpora, associated to a big distal urethral fistula. The urethral plate presented a fibrotic area under the hair constriction. There was no edema [Figure 1a and b]. The patient denied penile pain, including during spontaneous erections, and did not know about the PHT till the moment of our exam. The patient was long-haired as his mother had avowed to keep her son’s hair long till the cure of his hypospadias. This child had a long history of social deprivation.

After removing the hair tourniquet [Figure 1c], we verified normal patency of the balanic and proximal segments of the urethra, both easily catheterized with a 10 French catheter. We then opted to close the urethral fistula with a termino-terminal urethral anastomosis, after removing the fibrotic tissue under the constriction ring and debriding the two fibrotic extremities of the urethral fistula. The suture-line was protected with a darts vascularized flap, followed by glanuloplasty and approximation of the penile skin directly to the glans, after de-epithelialization of the proximal surface of the glans in the area of the sulcus.
The patient has now been followed up for 4 months, with satisfactory esthetic results [Figure 2] and no fistulae. The boy claims to have normal straight erections and urinates from a topic, despite irregularly shaped, meatus.

DISCUSSION

Less than 100 cases of PHT (penile hair strangulation, hair coil penile strangulation) have been published, but this syndrome may be responsible for a substantial proportion of penile pediatric trauma. Hair tourniquets may also be seen on toes, fingers and female genitalia: only about 1/3 involve the penis. Most patients are toddlers. The constricting hair strand normally lodges at the balanoprepucial sulcus (BPS) that provides a deep anatomical depression that holds the hair ring in place. The condition almost never occurs in uncircumcised boys, as the prepuce covers the BPS. Most cases are accidental, but some episodes of voluntary tying of hair around the penis in order to control urinary incontinence and some cases of abuse have been reported. Physiopathology includes progressive strangulation of the penis distal to the hair ring as the entrapped hair coil, initially loose, desiccates and becomes tighter, finally causing distal ischemia. PHT is typically painless, because of the slow progress of ischemia. Diagnosis is difficult because the constricting ring is inconspicuous (extremely thin and deeply hidden) and the disease is rare and not symptomatic. PHT is commoner among poor and neglected children: Poor hygiene may result in late diagnoses and referrals.

PHT may result in urethral transection (ventral or complete), glans ischemia and even distal penile amputation. Long delays in diagnosis may result in extensive fibrosis. In cases of urethral transection the treatment advocated by most authors is end-to-end urethral anastomosis, reinforced with a vascularized dartos flap. Complications (fistulae and stenosis) are relatively uncommon (16%), but the association with hypospadias may add to this risk. There is no data on timing for reconstructive surgery, but most authors advocate elective surgery just after resolution of secondary edema and cicatrization of superficial wounds. In cases of glans ischemia and separation, there are some reports of distal penile reimplantation.

There is only one other case report of PHT associated with hypospadias. This association occurs exclusively after surgery as a complete dorsal prepuce probably is as protective as the uncircumcised state. The association of PHT and hypospadias adds complexity to the repair as we deal with a post-operated scarred penis and a distal neo-urethra deprived from spongiosum. In our case, as there was no penile edema and superficial open wounds, the patient benefitted from the testosterone effect (local microvascular proliferation and more flexible tissues), and we opted for an immediate reconstructive procedure.

There are no available long-term results of the treatment of PHT complications. We were particularly worried about the possibility of abnormal erections, with no tumescence in the distal penis and glans and/or penile permanent hour-glass deformity. Thomas et al. reported a case of persistent hour-glass penile deformity and a case of permanent glans insensitivity in two children who presented without urethral transection and did not need penile reimplantation. This did not occur in our patient, who shows normal erections after 6 months follow-up.

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