CASE REPORT

Hair thread tourniquet syndrome in a male infant: a rare surgical emergency

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SUMMARY

Hair thread tourniquet syndrome (HTTS) is a rare surgical emergency that occurs when one or more appendages are acutely circumferentially strangulated by human hair. If left untreated it may induce prolonged ischaemia, resulting in tissue necrosis or autoamputation of the affected digit. It may involve the fingers, toes, penis or labia. It typically occurs in infants, but cases have also been reported in adults. Prompt recognition and treatment by complete removal of the constricting agent is crucial to preserve the affected tissue. We report a case of HTTS affecting the left middle toe of an 8-week-old male infant successfully treated by surgical release of the hair. The authors aim to raise awareness of HTTS among physicians, emergency doctors, paediatricians and surgeons, as prompt recognition and management prevents adverse outcomes and tissue necrosis.

and loupe magnification, the constricting cleft could be more thoroughly examined and the hair was removed. Two small stab incisions were made over the dorsolateral aspects of the toe to ensure complete division of the constriction. These incisions were planned to be dorsal to the neurovascular bundles and were continued to the depth of the bone. The wounds were irrigated and closed with 6.0 vicryl rapide sutures.

OUTCOME AND FOLLOW-UP

He was discharged the same day with a course of co-amoxiclav oral solution 125 mg/31 mg/5 mL.

One-week postdischarge, he was reviewed in the Plastic Surgery Dressings Clinic and the wound around his toe appeared to be healing well. Good perfusion of the distal portion of his toe was observed.

CASE PRESENTATION

An 8-week-old boy was brought to the emergency department with swelling and red discolouration of the left middle toe of 6 hours' duration. There was no history of trauma to the affected digit. His mother reported that he was becoming increasingly irritable, he appeared to be in pain and was crying continuously. He weighed 4.1 kg and his perinatal history was unremarkable. His immunisations were up to date. On examination of his left foot, a circumferential constriction around the left middle toe was noted, with reduced perfusion and a prolonged capillary refill time distally. On closer inspection, the toe appeared to be strangulated with a tourniquet formed of hair and cotton. Examination of the other toes showed no swelling or erythema. Attempts to fully remove the constricting hair using tissue forceps and a curved needle following the application of topical anaesthetic cream (EMLA) in the emergency department were unsuccessful. Hence, the child was referred to the Department of Plastic Surgery for further management.

TREATMENT

As it was uncertain that the hair had been removed completely, the decision was made to release the hair tourniquet surgically. The child was brought with his mother into an operating theatre. Local anaesthesia (lidocaine 1%) was injected as a digital block. After infiltration, the child was comforted by his mother. The area was then prepared with antiseptic and draped, with the child in his mother's lap. With the benefit of the operating lights

DISCUSSION

'Hair-Thread Tourniquet Syndrome' (HTTS) is defined as the ischaemic strangulation of an appendage caused by an encircling band of hair or thread. The term was introduced by Barton *et al*¹ following his experience of six infants, aged 12 days to 5 months, with hair and synthetic fibres strangulating an appendage. All six patients were treated by immediate removal of the constricting fibre and all cases healed without significant tissue loss. The condition was actually described earlier (Guillemieu in 1612) in a case affecting the glans penis.¹ In 1832 the *Lancet* published a case involving a hair intentionally wrapped around the penis of a 1-month-old infant by a 'vengeful nurse' recently dismissed from her employment.² It could be speculated that this, in fact, represented a case of HTTS. The tourniquet effect of the encircling material around the appendage causes lymphatic obstruction and oedema of the soft tissue.

The resulting swelling causes further obstruction of venous outflow and eventually arterial perfusion. Untreated, this will eventually lead to tissue necrosis and autoamputation of the affected digit.³

HTTS often affects the paediatric population, the most vulnerable period being in infancy. There is no gender predilection. The condition has also been described in adults with learning difficulties.⁴ The toes are the most commonly affected appendage (figure 1). The middle toe is involved most often. Cases affecting the earlobes, umbilicus and tongue have also been reported.⁵ HTTS affecting the toes is mostly caused by hair, while in the fingers they are

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Figure 1 Hair thread tourniquet. The toes are the most commonly affected appendage.

mostly caused by a loose thread from mittens. One possible risk factor is telogen effluvium, the diffuse hair shedding commonly experienced by almost 90% of women in the postpartum period. It is attributed to the variations in hormone levels (oestrogen, progesterone and prolactin), peaking in the first 6 months after birth.⁶ Hair is well-suited to become a tourniquet due to its high tensile strength and its ability to wrap around the tissue.⁷

Therefore, postpartum mothers, particularly those with long hair, should be counselled about this condition and the possibility of excessive hair loss after delivery. Trichotillomania, an impulse control disorder, characterised by the urge to pull out one's hair, has also been implicated as a rare risk factor.⁸ Most cases of HTTS arise accidentally and can be explained by the factors described. However, Klusmann and Lenard⁹ highlighted certain features in the presentation of five cases (inconsistent history, wrapping of the appendage with several knots) which raised the possibility of non-accidental injury. Recurrent presentations of

HTTS have also been suggested as a concerning feature.¹⁰ While clinicians must always be vigilant for non-accidental injury, it must be stressed that this is a rare cause of HTTS.

The diagnosis of HTTS is based on clinical findings, typically pain, discolouration, paraesthesia and swelling distal to a constricting band of hair. In the paediatric population, irritability may be the only initial presentation, leading to a delay in diagnosis. Therefore, a high index of clinical suspicion and careful physical examination are required. Delayed or missed diagnosis could lead to tissue necrosis, infection and autoamputation. Strangling injury to the penis by hair may result in devastating complications such as urethracutaneous fistula and glandular amputation.

Therefore, all patients presenting with penile pain and swelling should be examined for HTTS.¹⁰ In young infants, congenital constriction band syndrome should also be considered in the differential diagnosis. In addition, digital annular constriction affecting a toe, especially in one of African descent, should raise the suspicion of the condition *ainhum* (*dactylosis spontanea*) or *pseudo-ainhum*, of which hair tourniquet syndrome is a subgroup.⁹

Acute management of HTTS involves administration of adequate analgesia and relief of the acute strangulation through removal of all constricting bands and monitoring of the neurovascular status of the digit for signs of ischaemia (*figure 2*). Often this is possible in the emergency department. Assessment in a room with good lighting and the use of fine scissors and forceps under magnification are recommended. However, in some cases it is not possible to remove the constricting material, and surgical exploration for complete release will be required.¹¹ Some studies have advocated use of depilatory creams containing thioglycolate and calcium hydroxide or sodium hydroxide to dissolve the hair atraumatically.^{12 13} The depilatory agent is applied for 10 min, thus allowing the thioglycolate to break down the

Figure 2 An algorithm for the management of hair thread tourniquet (HTS) syndrome.

disulphide bonds of keratin, thus releasing the constriction. Relative contraindications to its use include broken skin or a history of allergy. However, this method will not be successful if the tourniquet is formed of cotton, polyester or rayon.¹⁴

Surgery can often be undertaken under local anaesthesia, even in infants. The child can be brought to the operating theatre and held by the parent in their lap. Following infiltration of local anaesthesia, the infant can receive a feed for comfort often, subsequently becoming sleepy. The area can be prepared and draped, with the child in the parent's lap. If local anaesthesia is not felt to be appropriate, general anaesthesia should be used. Serour and Gorenstein reviewed the surgical techniques described to deal with HTTS. They advocated a short but deep incision made in the longitudinal axis of the digit on the dorsal side of the toe until the bone is reached.¹⁵ This technique secures the complete release of the strangulation by dividing the thread while avoiding injury to the neurovascular structures of the toe. The cleft is explored and any thread and hair removed. García-Mata and Hidalgo-Ovejero¹⁶ proposed performing two dorsal paratendinous incisions, deep to the bone, to completely release

the strangulation. A course of antibiotics postoperatively is usually advised.¹⁷

Prognosis depends on the degree and duration of ischaemia and therefore ranges from full recovery to amputation of the affected digit. According to Kurup *et al*,¹⁸ involvement of the toes has a better prognosis compared with other appendages.

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Learning points

- Hair thread tourniquet syndrome typically presents with pain, discolouration, paraesthesia and swelling in an affected appendage, distal to a constricting band of hair.
- Children presenting with irritability and no other signs of systemic illness should have their digits examined for signs of strangulation.
- It is an emergency condition that induces progressive oedema, ischaemia and tissue necrosis, and can lead to autoamputation of the affected appendage.
- Prompt recognition of 'Hair Thread Tourniquet Syndrome' and complete removal of the offending constricting agent are required.
- Depilatory creams may be useful as an adjunct to attempting removal of a hair tourniquet in the emergency department.
- If it is not possible to remove the constricting ligature, or if there is doubt as to whether the constriction has been completely relieved, the patient should be referred for surgical exploration. This can be undertaken under general or local anaesthesia and involves making one or two incisions dorsal to the neurovascular bundles.

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