



Review article

Hair Tourniquet Syndrome, A missed diagnosis: Review of Literature

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ABSTRACT

Hair tourniquet syndrome is a rare yet potentially devastating entity in children. Hair-thread tourniquet syndrome involves fibres of hair or thread wrapped around an appendage (toe, finger, clitoris, penis etc.) producing tissue necrosis. It is frequently missed or misdiagnosed leading to delayed diagnosis resulting in gangrene of toe or finger. Tourniquet syndrome of penis can present with total or partial amputation or as chronic urethrocuteaneous fistula. Its frequent association with maternal telogen effluvium makes it a potentially a preventable condition. Treating physician must be aware about potential of child abuse as a cause of hair tourniquet syndrome.

KEYWORDS: Toe tourniquet syndrome, Hair tourniquet syndrome, Acquired constriction ring syndrome, Genital tourniquet syndrome

INTRODUCTION

Hair-thread tourniquet syndrome involves fibres of hair or thread wrapped around an appendage (toe, finger, clitoris, penis etc.) producing tissue necrosis. The first cases involving the hair strangulation of appendages were reported in 1965 by Alpert and colleagues. However it was Quinn who first created the term "toe tourniquet syndrome" in 1971 which described the effect of hair wrapping around the toes leading to swelling. Barton et al [1] coined the term hair-thread tourniquet syndrome in 1988. It is now being referred to as hair tourniquet syndrome or acquired constriction ring syndrome.

Tourniquet syndrome occurs infrequently, and the incidence is not known. Claudet et al [2] described the epidemiology of a group of children admitted to a paediatric emergency department for tourniquet syndrome over a six year period: the mean age was 5.5 months among the 57 patients, and Barton et al reviewed 32 papers published before 1988 and collected 66 cases suffering from tourniquet syndrome: the median ages of patients were four months, two years, and three weeks for toe, external genitalia and finger groups respectively. These two papers represented the largest case series on hair tourniquet syndrome in the literatures. Claudet et al [2] found that majority of the tourniquet syndrome cases (95%)

involved toes while the remaining common sites were penis and labial majora. Barton found that 28 cases (43%) involved toes, 22 cases (33%) involved the external genitalia among which great majority (95%) affecting the penis whereas only one case affecting female genitalia, and 16 cases (24%) involved the fingers. Claudet's study, noting that hair responsible for 95% of all cases; and Barton's study, noting that hair responsible for 79% of toe cases, 95% for genital cases, and thread responsible for 80% of the finger cases.

PRESENTATION

Characteristically the child will present with obvious irritation. On examination the affected toe(s) will be swollen, congested, and oedematous. If the texture of the constricting material is fine and occult, such as hair, initial presentation can only be swelling and redness of the affected part of the body. Lymphoedema produced impedes venous drainage, causing more oedema, and eventually compromises arterial circulation to the tissue leading to tissue gangrene.[3] The hair fiber may even cut through the oedematous skin, then deeply embeds in the subcutaneous tissue, and subsequently epithelializes in the skin, making the hair thread obscured and further delaying the diagnosis.

Hair Tourniquet Syndrome and Maternal hair loss

It is suggested that women going through the postpartum period following pregnancy can experience severe hair loss. This is known as telogen effluvium. Pregnancy is a major hormonal process in the body and the cessation causes distress to hair growth, and therefore the mother would experience increased hair loss (>100 hairs per day). Such hair loss can be induced over the postpartum months 4 - 6 before eventually transitioning back to normal. It occurs in roughly 90% of post-partum mothers. As the peak incidence for toe tourniquet syndrome was at around four months of age, correlation with the period of maternal post-partum alopecia was

postulated.[4] Human hair, stretched when wets, and contracted and tightened when dries, is special in causing tourniquet effect as it is extremely thin with a tensile strength of greater than 29,000 lbs/square inch[5]. While string and hair are equally as dangerous, hair is often more worse because it can wrap around an infant's toe so tightly that the skin folds over the hair. Baby cloths such as mittens and single piece jumpsuits may cause accumulation of hairs and may pose increase risk of hair tourniquet syndrome.

Hair Tourniquet syndrome and Child abuse

The earliest account of hair strangulation was reported in 1832 by Dr G who removed a hair band around the penis in a one-month-old infant.[6] This was in fact a child abuse case wherein the hair thread was placed by a revengeful nurse who had been recently dismissed from her duty. A case series by Klusmann et al [7] ascribed all five tourniquet syndrome cases to some form of child abuse.

When a case of hair tourniquet syndrome in a young child is encountered, the lack of reasonable explanation and the presence of meticulous wrapping, if any, shall make non-accidental injury a possible cause. Professor Sir Samuel Roy Meadow,[8] who first introduced the term Munchausen Syndrome by Proxy, gave some comments on tourniquet syndrome in 2004: "The boundaries of child abuse are more and more extensive and abusive acts are recognized which are beyond the imagination of most people. It is correct to re-examine the cause of any unusual or strange condition and enquire whether it might be the result of deliberate harm or neglect".

In a survey of healthcare and child protective services in Miami, 83% of child welfare workers and 45% of public health nurses misinterpreted this as intentional injury.[9] Most cases of involvement of the fingers and toes tend to be accidental while constriction around the genitals is frequently non-accidental in nature.[10] Child abuse has to be seriously considered for most if

not all tourniquet cases. A through history and circumstance of presentation has to be looked into carefully as incorrect accusations of abuse may cause as much harm as failure to recognize.

Genital Tourniquet Syndrome

Genital tourniquet injury is rare entity and has been reported following application of numerous foreign objects, including rings, rubber bands bottle pipe and thread. Reports of hair tourniquet syndrome of genitals have been reported in young. Contrary to digital variety the genital cases are usually seen in relatively older children ranging from 4–11 years. This age corresponds to the Sigmund Freud's phallic (or clitoral) stage of psychosexual development when kids start toying with their genitalia out of curiosity. The application of a tourniquet around genitalia may be a possible reflection of this psychosexual tendency.

Genital tourniquet syndrome usually involves penis though there are reports of clitoris [11] and labia minora [12] being affected in girls. Strangulation of penis, by an ancient folklore, was used to ward off evil spirits. In more recent times it has been reported to have been used to prevent enuresis or nocturnal emission. [13]

Circumcision is an important predisposing factor, since it appears that the condition is much more common in circumcised boys. [14] Presentation in the acute state is usual, with swelling, erythema, circumferential constriction and distal edema [15] with little discomfort to the child. [16] If the tourniquet is not removed early, there may be progression to skin infection and ulceration, but removal of the constriction at this stage prevents long-term complications. In cases that are not recognized early, the dorsal neurovascular bundle may be transected, leading to the loss of sensation over the glans penis. The corpus spongiosum may become transected, leading to urethrocutaneous fistulation. The corpora cavernosa may also become transected, leading to partial or total amputation of the penis distal to the tourniquet

[14] Young boys especially circumcised presenting with penile or glanular swelling, purulent discharge or urine leaking from coronal sulcus, or a gangrenous glans must be suspected of having genital tourniquet syndrome.

Bashir and EL-Barbary[17] categorized penile strangulation as: Grade 0: Constriction of skin without urethral injury, Grade 1: Partial division of corpus spongiosum with an urethrocutaneous fistula, Grade 2: Complete division of the corpus spongiosum and constriction of the corpus cavernosum and Grade 3: gangrene, necrosis and, complete amputation of the glans.

Treatment is aimed at relieving the constriction by cutting or unwinding the hair. In cases where there is partial amputation or injury to urethra single stage repair is performed with catheter stenting. [18]

Immediate decompression remains the main treatment. A thorough examination of all toes and fingers and genitalia must be performed to avoid missing simultaneous involvement. In initial stages associated with swelling and erythema attempts at identifying the constricting material under magnification is made and unrolling or cutting of hair is done with needle or fine scissor. If unable to pass scissor below the hair or there is risk of damaging the skin and swelling is less attempt to dissolve hair with use of depilator can be made [19], however this would not be effective in case of synthetic fibres or if hair has penetrated the skin.

In advanced stages when swelling is profuse or skin has breached, then it is advisable to carry out urgent complete decompression in theatre preferably under general anesthetic. Adequate light and magnification will further aid the search for constricting agent which may be lying buried deeply inside the subcutaneous tissues. The skin may re-epithelialize over the buried hair making the exploration further difficult.

Serour and Gorenstein[20] have described in a thorough manner the technique they have employed to remove hairs or fibres in cases of toe tourniquet syndrome. The first step would involve disinfecting the area with povidone-iodine or chlorhexidine solution before injecting a local anesthetic such as lignocaine to either side of the toe. Afterwards, a short but deep incision is made in the longitudinal direction on the dorsal side of the toe until the bone is reached. They believe that doing this can avoid any injuries made to the neurovascular bundles that sit on the lateral and medial aspects of the toe as well the lymph vessels on the plantar side. Then the hair or thread is removed using a forceps and a dressing of antibacterial cream and petroleum jelly should then be applied to the affected area to aid wound healing. It is vital to examine the tourniquet for presence of a well formed knot as it would strongly suggest presence of non accidental injury.

Reported complications are tissue loss, flexion deformities, and amputation.[21] Postoperative complications include rotation of the soft tissue around the tip of the toe, causing the nail to shift in a medial direction.[22] This can be avoided by a single tacking stitch after decompressing the digit.

Counseling of postpartum mother is imperative and crucial. It is important to educate her to perform through regular checks of limbs to ensure that no hairs are entangled around fingers and toes of the baby. Mittens, single piece jumpsuits and clothing covering fingers and toes should be regularly checked for presence of loose hairs and should be washed inside out. Mothers should seek medical attention immediately if the first sign of entrapment or circulation compromise is noticed or suspected.

CONCLUSION

Hair tourniquet syndrome is rare but potentially devastating entity in children. Every parent and physician attending to children must be aware about this phenomenon to avoid misdiagnosing or delay in diagnosing which could lead to loss of

appendage. Any child presenting with swelling and erythema of digits must be evaluated for hair tourniquet syndrome under magnification or surgical exploration. Treatment is immediate decompression of digit with vertical slit and removal of constricting material. Its increasing association with maternal telogen effluvium makes it necessary to educate mothers about the precautions to be taken. All cases of hair tourniquet syndrome must be carefully investigated for probable child abuse or neglect and social services informed to take necessary action.

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