

Hair-Thread Tourniquet Syndrome in an Infant With Bony Erosion

A Case Report, Literature Review, and Meta-analysis

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Abstract: Hair-thread tourniquet syndrome is a rare condition where appendages are strangulated by an encircling strand of hair, a thread, or a fiber. The condition usually occurs in very young patients in the first few months of life. We present a unique case of a 3-month-old baby girl with hair-thread tourniquet syndrome in whom a hair cheese-wired through the skin and soft tissue of the toe and caused bony erosion of the underlying phalanx. An extensive literature review and meta-analysis of the topic are also presented.

Key Words: hair, thread, toe, finger, penile, clitoris, tourniquet syndrome

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Hair-thread tourniquet syndrome is a rare condition that occurs mostly in babies, just a few months old. It occurs when a strand of hair or occasionally a piece of thread or fiber encircles an appendage, causing partial or total obstruction to circulation. Cases have been described involving the toes (toe tourniquet syndrome), fingers, genitalia, uvula, and neck. Although most cases are felt to be accidental, nonaccidental injury must be considered in a minority of cases.

The offending fiber cuts through the skin, making it difficult to detect. Irritability may be the only presenting symptom; thus, a high index of suspicion is required. Appropriate treatment is prompt removal of the constricting fiber. This can usually be achieved under direct vision; however, the hair can be buried and obscured by reepithelialization of the skin, making complete removal difficult to ascertain.

From our extensive literature review and meta-analysis, more than 200 cases have been reported on this syndrome.

We present an unusual case of hair-thread tourniquet syndrome where a hair cheese-wired through the skin and soft

tissue to cause bony erosion of the underlying phalanx of a toe.

CASE REPORT

A 3-month-old baby girl was referred to our unit from the emergency department, with a history of irritability and a red swollen right middle (third) toe, which failed to resolve 3 days after removal of a hair tourniquet (in the emergency department of the referring hospital).

Careful examination in our own emergency department with loupe magnification showed intact skin on the toe and no further evidence of a residual hair tourniquet. A course of antibiotics was prescribed for cellulitis, and improvement was noted on review in our outpatient department at 1 week.

Six weeks later, the patient represented to the outpatient department, with recurrent swelling and redness of the toe. A small erosion with a slight discharge was identified over the dorsal distal interphalangeal joint, and a further course of antibiotic was prescribed. However the eroded area deteriorated, ulcerated, and further close examination in the clinic revealed a hair buried beneath the area of skin breakdown (Fig. 1). X-ray at the time showed an erosion of the middle phalanx of the toe (Fig. 2). Exploration and complete removal of the hair tourniquet was performed under general anesthesia. Intraoperatively, it was noted that the skin had completely reepithelialized over several strands of hair, which were buried partly within the bone of the middle phalanx. The hairs were removed completely, and the bone was curetted. A full and uneventful postoperative recovery ensued.

METHODS

A MEDLINE search was performed through PubMed using the keyword *tourniquet syndrome* in December 2005 (no year limit and all languages included). Relevant articles were obtained, and references from each of these articles were further searched for relevant articles. A total of 86 articles were reviewed (of which 77 were case reports or series). There were a total of 210 reported cases from which data were collated and analyzed.^{1–59,61–67,69–86}

RESULTS

From our extensive review of literature and meta-analysis of this syndrome, there are 3 major series published in the past. Haddad³³ published a series on penile hair strangulation with 60

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FIGURE 1. Right third toe with swelling, erythema, and a small area of skin erosion.



FIGURE 2. X-ray of the toes showing swelling of the soft tissue and erosion of the middle phalanx over the distal-lateral aspect (see arrow).

cases in 1982; then Barton et al,¹³ who coined the term *hair thread tourniquet syndrome* (HTTS), published a series with 66 cases involving fingers, toes, and external genitalia in 1988. The last major series was published in 2005 by Golshesky et al,⁶ with 90 cases. The cases from these series were compiled, and care has been taken to avoid double recording from overlapping cases within these series.

In total, our literature review yielded 210 reported cases, with 259 appendages involved. We found that penile involvement occurred in 44.2%, toes in 40.4%, fingers in 8.57%, and other sites (female external genitalia, uvula, neck) in 6.83% of cases. Like previous series, this one is best further subdivided into 3 categories, toes, fingers, and external genitalia, for statistical purposes (Tables 1 and 2).

The patients with toe injuries ranged from 2 weeks to 84 years of age (median, 4 months). The 84-year-old Alzhei-

mer's patient is the only one recorded in the toe cases beyond the age of 6 years old. Of these cases, 72 (85%) were caused by hair, 12 (14.11%) were by loose thread, and in 1 case, both hair and thread were involved. The third toe was most frequently affected, with 41 cases (31.5%), followed by fourth toe 31 (23.8%), second toe 22 (16.9%), little toe 3 (2.3%), and 2 (1.5%) in the big toe. However, there were 31 (23.8%) cases where the exact toe was not specified. Multitoe involvement was reported in 33 out of 85 cases, with 2 toes involved in 20 (23.5%) cases, 3 toes with 11 (12.9%), and 4 toes in 1 (1.17%) case. Three patients had complications including distal segment rotation and 2 with amputation.

Fingers were affected in 18 cases, with 22 affected digits. The children ranged between 4 days and 19 months old (median, 3 weeks), and the majority (>80%) were younger than 2 months old. Thirteen cases (83.3%) were caused by thread or fiber from clothing items, usually mittens. The index finger was the most frequently affected, with 10 (45.5%) cases, followed by the middle finger with 8 (36.3%) cases, the thumb with 3 cases, and the ring finger in 1 case. There were no reported cases involving the little finger. Multiple digits were involved in 3 out of 18 cases, of which 2 digits were involved in 2 cases and 3 digits in 1 case. The complication rate at 54.4% is high and included amputation in 4 cases, gangrene in 5 cases, and soft tissue loss in 3 cases.

There were 103 cases involving external genitalia predominantly involving the penis (93 cases), followed by the clitoris (7 cases), labia (2 cases), and mons pubis (1 case). For penile strangulation, the age ranged from 2 days to 77 years (median, 2 years). In children, the age ranged from 2 days to 14 years, while 2 adult males were 69 and 77 years old. In females, the age ranged from 5 to 14 years (median, 8 years). Almost all cases (97%) were caused by hair strangulation. The complication rate is high at 46.6% (Table 3).

There were 2 reported cases of uvula strangulation and another 2 of neck strangulation by hair to complete the series.

DISCUSSION

The term *toe tourniquet syndrome* was first introduced by Quinn⁴⁴ in 1971. Nearly 2 decades later, Barton et al¹³ coined the term *hair-thread tourniquet syndrome*, which broadly covers this condition where a strand of hair or a thread or fiber encircling an appendage causes ischemic strangulation.

This condition has been recognized since the 1600s, when strangulating hair was reported around the penis (by Guillimeau⁸²) (quoted from Haddad,³³ Thomas et al,⁷⁰ and Guillimeau⁸²). Since then, sporadic reports appear in the literature.³ Among the early references available were Gaultier in 1755 (quoted from Mackey et al² and Gaultier⁸¹) and Dr. G in *The Lancet* in 1832 (of penile strangulation).⁵³

These syndromes were normally described in very young patients. The finger strangulation mostly occurred in their first couple months of life. In slightly older infants, the toes are commonly affected. Penile tourniquet is more common in patients around 2 years old.^{13,33} The labial or clitoral

TABLE 1. Results^{1-59,61-67,69-86}

	Grand Total	Appendage							
		Toe	Finger	Penis	External Genitalia			Others	
					Clitoris	Labia	Mons Pubis	Uvula	Neck
Total reported cases (%)	210 (100%)	85 (40.4%)	18 (8.57%)	93 (44.2%)	7 (3.3%)	2 (0.95%)	1 (0.47%)	2 (0.95%)	2 (0.95%)
Total appendage (%)	259 (100%)	130 (50.2%)	22 (8.5%)	—	103	(39.7%)	—	4	(1.5%)
Multiple appendages cases	36 of 210	33 out of 85	3 out of 22	—	—	—	—	—	—
Reported age range (median)	4 days–84 years (6 months)	2 weeks–84 years (4 months)	4 days–19 months (3 weeks)	2 days–77 years (2 years)	5 years–14 years (8 years)	13 years–14 years	7 years	3 months–13 months	11 months–27 months
Average age	2 years 11 months (1 year 9 months)*	5.3 months [†]	2.9 months	2 years 10 months [‡]	8 years 10 months	13 years 6 months	7 years	8 months	19 months
Offending fiber									
Hair	179 (85.2%)	72 (84.68%)	3 (16.6%)	91 (97.8%)	6 (85.7%)	2 (100%)	1 (100%)	2 (100%)	2 (100%)
Thread	29 (13.8%)	12 (14.11%)	15 (83.3%)	1 (1.07%)	1 (14.2%)	0	0	0	0
Both	2 (1%)	1 (1.17%)	0	1 (1.07%)	0	0	0	0	0
Gender									
Male	144 (68.5%)	44	5	93	0	0	0	1	1
Female	44 (20.9%)	28	4	0	7	2	1	1	1
Not specified	22 (10.5%)	13	9	—	—	—	—	—	—
Complications		3 (2.3%)	12 (54.5%)	46 (52.6%)	1 (14.2%)	1 (50%)	0	1 (50%)	0

*Excluding 69-, 77-, and 84-year-old patients.

†Excluding an 84-year-old Alzheimer's woman.

‡Excluding 69-year-old and 77-year-old man.

TABLE 2. Breakdown of Toe and Finger Cases

	Toe	Finger
Big toe/thumb	2 (1.5%)	3 (13.6%)
Second toe/index finger	22 (16.9%)	10 (45.4%)
Third toe/middle finger	41 (31.5%)	9 (36.3%)
Fourth toe/ring finger	31 (23.8%)	1 (4.5%)
Fifth toe/little finger	3 (2.3%)	0
Not specified	31 (23.8%)	—
Multitoe/digit involvement	33 of 85 cases	2 of 18 cases
Two toes/digits	20 (23.5%)	2 (11.1%)
Three toes/digits	11 (12.9%)	1 (5.55%)
Four toes/digits	1 (1.17%)	0

strangulation is described in much older children (5–14 years).^{3,13} There are occasional cases reported in elderly patients well past their sixth decade.^{7,33}

Early recognition of this condition is important and requires a high index of suspicion when no other causes are found in an irritable infant. Delayed diagnosis may result in loss of appendages in some cases.^{1,3,13,17,19,23,24,33}

If the hair or thread has not been removed, the tourniquet effect causes lymphatic obstruction, edema of tissue, and

TABLE 3. Complication From Genitalia Appendages Cases

Penile cases		
Complete amputation	4	4.3
Partial amputation	15	16.1
Urethral fistula	24	25.8
Wasp-waist deformity	4	4.3
Urethral fistula and partial amputation	2	2.15
Total no. complications	49	52.60%
Female external genitalia cases		
Partial amputation	1	Clitoris
Complete amputation	1	Clitoris
Tissue loss	1	Labia

swelling. A vicious circle is started as the more the tissues swell, the tighter the tourniquet becomes. Secondary venous congestion follows, and ultimately arterial perfusion is impaired.^{2,9,14,15,23} This process may progress rapidly over hours or more insidiously over months.^{2,18}

In some instances, the fiber can cut through and become buried under the skin, making detection at examination difficult. Re-epithelialization of the skin over the fiber may also occur, thence obscuring the offending fiber altogether.^{18,23}

Etiology

The etiology of these conditions is believed to be accidental in most cases. The precise mechanism of entanglement is variable. Many hypotheses have been proposed to explain the phenomenon, and most authors believe it happens purely by chance with baby's digital movements within the loose fabric of clothing such as mittens or socks.^{2,44} Quinn⁴⁴ in 1971 has postulated the entity is caused by combination of circumstances such as type of clothing (especially leotard-type garments), which accumulate foreign material, and variable response to plantar reflex in infants.

Several authors have suggested that this type of clothing be turned inside out before laundering, which reduces or eliminates the chance of human hairs or thread being harbored after repeated washing.⁶⁷

In the case of toes and genital appendages, they are mostly caused by hair, while in the fingers, they are mostly caused by a loose thread from mittens.^{2,13,67}

Alpert et al⁴⁵ in 1965 proposed that the quality of hair makes it a particularly effective constricting agent, ie, human hair is quite stretchable when wet and contracts when dry.³⁹

Strahlman¹² has described a direct association between maternal hair loss (telogen effluvium, which occurs typically between 2 to 6 months postpartum) and the occurrence of hair-thread tourniquet syndrome.

Haddad,³³ in his detailed review of penile strangulation, has divided the etiology into 4 groups. They are accidental, incidental, intentional, and undetermined cause. He also mentions 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.

Nonaccidental cause should also be considered carefully in certain cases as mistake and misjudgment have been recorded in the past. There are several reports in the literature where the diagnosis of child abuse has been overturned.^{48,54,73}

Historical literature has described cases where the hair or thread was intentionally placed to elicit injury.⁵³ In some cultures, wrapping hair around toes or fingers of the newborn is believed to ward off evil spirits.^{33,45} In other cultures, wrapping of a hair or ligature around the penis is used to prevent nocturnal enuresis.^{33,69,70}

In the modern literature, the nonaccidental cases were almost unheard of until Klusmann and Lenard⁸ reported 4 cases, which they strongly believed were that of child abuse. They argue that a lack of explanation by the parents (all cases), involvement of multiple digits, and multiple strands of hair with tight knots present are indicators of nonaccidental injury. We feel that using these facts as evidence for nonaccidental injury is open to discussion and debate. As we can see from previous reports, involvement of multiple digits and the discovery of knots in the fiber are not uncommon, and in most cases the parents can give no explanation.

However, suspicion for nonaccidental injury should increase when the strangulation material is found with knot(s) tied in it.³⁴ General consensus for the past few decades is that hair-thread tourniquet syndrome is mostly an accidental injury.

Nonetheless, the possibility of child abuse cannot be ruled out automatically once the diagnosis is made, and careful history taking is necessary.⁶

A survey of health care workers by Biehler et al³¹ found that a child welfare worker will interpret this syndrome as child abuse and will make referral for suspected child abuse (89%) more frequently than a physician (53%–56%) or public health nurse (48%). They later suggested that hair-thread tourniquet syndrome be published in the child abuse literature.

Care should be taken and senior physicians must be consulted prior to making an allegation of nonaccidental injury because it may have adverse consequences for the parents and the treating physicians, as well as the child.

Several authors have suggested that in some cases where older children were involved, especially with genital appendages, that the injury may have been self-inflicted for experimentation.^{5,6,14}

Among other differential diagnoses for a digital annular constriction should include ainhum (dactylolysis spontanea),^{60,68} pseudoainhum,³³ pachonychia congenita, pityriasis rubra pilaris, mal de Meleda, and also congenital constriction band.²⁹

In the case we report, the bony erosion on x-ray and clinical picture of swollen inflamed digit mimicked osteomyelitis.

Management

Complete removal of the encircling fiber is essential to restore the circulation. This can be done with the aid of a magnifying glass in the accident and emergency setting^{2,7} or in the operating theater. If a complete removal cannot be ascertained, surgical exploration is mandatory. This can be done under local anesthetic¹⁰ or under general anesthesia. A simple longitudinal incision along the digit (either middorsal or lateral aspect of the toe or the finger) down to bone is safe and effective to completely remove the constriction.^{2,10} There are more than a dozen reported cases where the strangulation was not fully released in the first attempt and surgical intervention was required to completely remove the offending fiber.^{10,11,13,16,18,26,42}

Kerry and Chapman⁴⁷ have made the same point that in severely edematous tissue, it is difficult to determine whether the constriction has been removed completely. They have suggested that surgical incision be made in the lateral aspect of the finger/toe down to the bone. In case of penile strangulation, incision can be made in the inferolateral aspect between corpus cavernosum and corpus spongiosum (should be done preferably by a specialist).

Mack et al³⁹ had reported 2 cases of toe tourniquet syndrome, of which one was complicated with rotation of the tip of a digit, though it healed without tissue loss. They had suggested placing a single tacking suture to prevent rerotation after the offending fiber had been released.

The patient should be reviewed regularly after the encircling fiber has been removed to confirm complete resolution of strangulation.

In extreme scenario, the risk of amputation or resection of devitalized tissue may be necessary.

CONCLUSIONS

Hair-thread tourniquet syndrome is an uncommon entity that has gained a lot of recognition in the past decade. Many case reports exist describing this entity involving differing appendages. Early diagnosis and prompt treatment can prevent loss of appendages and save the patient from unwanted complications, which occur in up to 50% of cases. We believe our case is the first in the literature to report bony erosion. This was visible on x-ray, and we suggest a plain film may be a useful investigation to detect deep tourniquets causing persistent strangulation where the surface appearance is normal and deceptive. We also present this case to further educate ourselves and young surgeons on the topic and create awareness of the existence of the syndrome for all who might encounter this clinical entity.

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