

Commonly Misdiagnosed Rare Condition—Toe Tourniquet Syndrome—Literature Review

Muhammad Ali Hussain^{1*}, Tonkid Taneerananon¹, Simon Wharton²

¹The Plastic and Reconstructive Surgery Department, The Canberra Hospital, Canberra, Australia; ²Plastic Surgery Department, Russells Hall Hospital, West Midlands, UK.
Email: *dralihussain76@yahoo.com.au

Received November 15th, 2011; revised December 30th, 2011; accepted January 12th, 2012

ABSTRACT

Toe Tourniquet Syndrome (TTS) is a rare yet potentially devastating, acquired defect, which is repeatedly misdiagnosed in the United Kingdom and abroad. Due to the relatively infrequent presentation, it can be easily missed. Toe tourniquet syndrome is strangulation of the toes by what is usually hair, although synthetic fibres have also been described. It is considered a surgical emergency as left untreated, the tissues of the toe can become necrotic and the patient would lose that appendage. In this paper, we have done a literature review and written the article with illustrations which would explain this repeatedly misdiagnosed condition very thoroughly to all the clinicians dealing with this rare problem.

Keywords: Toe Tourniquet Syndrome; Repeatedly Misdiagnosed Condition; Review

1. Introduction

Toe Tourniquet Syndrome (TTS) is a rare yet potentially devastating, acquired defect, which is repeatedly misdiagnosed in the United Kingdom and abroad. Due to the relatively infrequent presentation, it can be easily missed. Toe tourniquet syndrome is strangulation of the toes by what is usually hair, although synthetic fibres have also been described [1]. It is considered a surgical emergency as left untreated, the tissues of the toe can become necrotic and the patient would lose that appendage.

TTS has been reported to affect patients from as young as 2 weeks to infants aged under 6 years old. However one elderly patient aged 84 years who suffered from Alzheimer's disease has been the only exception to this [1,2]. This syndrome has been described primarily affecting the toes of young children averaging around 4 months old. In a review of 66 cases, toes were affected in 43%; fingers 24%; and hair-thread tourniquet in the genital regions 33% [3]. Hair was the most common offending agent affecting toes, while thread or fibre, normally attributed to mittens, was more common in the situations where fingers were concerned [1,4].

2. Background

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.

The reasons for hair winding so tightly around the toes are still unknown. It is believed that when hair is moist, it is suppler whereas once it becomes dried up; it can condense and cause further constriction [5]. Also, due to the thin nature and high tensile strength of hair, it effectively becomes a garrote, slicing through the skin during this transformation [6]. This is exacerbated by the consequential oedema arising from the tourniquet.

3. Presentation

Characteristically the child will present with obvious irritation. On examination the affected toe(s) will be swollen, congested, and oedematous. In more serious cases, the hair or thread will have cut through the skin and into the soft tissue (**Figure 1**). It can be hard to find as infants have such small appendages and it can easily be masked by skin folds and oedema. Once the skin is cut, skin epithelialisation can occur which buries the offending strand of hair or thread, also making it difficult to diagnose.

4. Risk Factor

4.1. Mother

4.1.1. Telogen Effluvium

It is suggested that women going through the postpartum period following pregnancy can experience severe hair loss [3]. This is known as telogen effluvium. Pregnancy is

*Corresponding author.

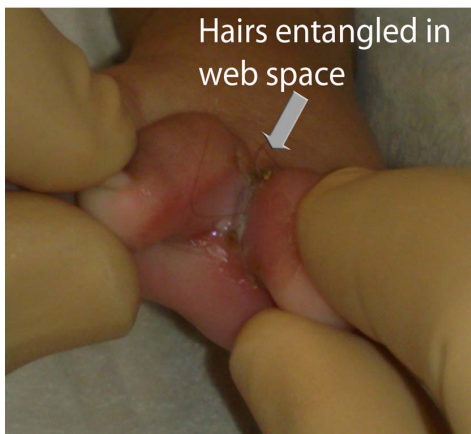


Figure 1. Hairs entangled in web space.

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.

4.1.2. Neglect

Toe-tourniquet syndrome is considered a rare case and on presentation in hospital it should never be ruled out that there may be possible neglect or abuse of the child. However it can be easily misinterpreted and strangulation of the toes and fingers by large is more likely to be unintentional whilst a constriction within the genitalia is more likely to be a non-accidental feature [7]. Neglect in washing the child properly and checking the appendages whilst bathing can also be considered. New mothers should be educated in excessive hair loss and make sure none of the hairs become trapped around the fingers and toes and ideally all clothes covering these areas should be turned inside out to make sure no hairs are caught.

4.2. Baby

The age of the patient is also significant. Babies regularly play with the toes and also they are unable to communicate clearly, with crying and irritability being the only signs, and these are attributed as usual symptoms in patients of this age, therefore the TTS is only brought to the attention of the parent when the appendage is swollen and ischaemic [8]. Barton has also suggested that a contributing factor is that some infants have variable plantar reflexes [1].

4.3. Season

It is suggested that this phenomena is reported more frequently in the summer months compared to winter. Such reasoning could be due to the fact that the infant is less likely to wear clothing covering the toes. Kids usually play

with toes when not wearing baby grows. Whilst walking around, the child could easily pick up the hairs without noticing and eventually find that the hairs wind tighter and tighter around the toes, causing swelling and pain. Garcia-Mata and Hidalgo-Ovejero described how due to the hot weather during the summer months, frequent bathing, whether at home or public swimming pools can increase the risk of picking up hair remnants [8].

4.4. Carpet in the Home

Another risk factor is having a home with fitted carpets. Ideally on a laminated or tiled floor, hairs are more easily noticed and swept up whereas in a home with carpets, the hairs are much more difficult to find. Without a decent vacuum, the hairs may not necessarily get swept up efficiently following post-partum.

4.5. Race

Race may affect this condition as well. Fair-coloured hair on fair-skinned infants is harder to locate than a dark coloured hair. Although cases of both have been reported, in cases where fair coloured hair is found on fair skin infants, the cause of the child's irritation on presentation is much harder to diagnose.

5. Treatment

The most common and effective treatment is to remove all the hair or threads surrounding the toe by surgery, relieving it of strangulation and providing immediate re-circulation of the blood and lymph vessels within the area. If not all hair or fibres are removed from the area, symptoms may not subside and further intervention is needed. There have been many cases reported where more than one strand has been pulled from the affected areas [8]. Chemical hair removal formulae such as depilatory creams have been suggested as a way to help dissolve the hair [9]; however this would not work on synthetic fibres or hair that had already cut deep past the skin level [10].

Surgical Technique

Serour and Gorenstein have described in a thorough manner the technique they have employed to remove hairs or fibres in cases of toe tourniquet syndrome [10]. They have reviewed the surgical techniques believed to have been in use between 1990 and 2001 on most TTS presenting patients. The first step would involve disinfecting the area with povidone-iodine or chlorhexidine solution before injecting a local anaesthetic 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.

Once this incision had been successfully made, the transected hair or fibres will become more superficial and in most cases will be easily removable with forceps (**Figures 2-5**). A dressing of antibacterial cream and petroleum jelly should then be applied to the affected area to aid wound healing.



Figure 2. Use of micro instruments to remove the hairs.



Figure 3. Postoperative pictures after removal of entangled hairs—planter view.

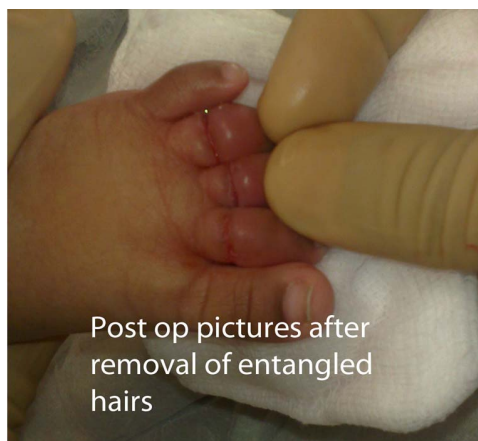


Figure 4. Postoperative pictures after removal of entangled hairs.



Figure 5. Removed mother's hair.

If the hair has cut through the skin with skin epithelisation occurring, it becomes difficult to ascertain whether all hair and fibres have been removed. In this case surgical intervention under general anaesthetic is needed [11]. Haene & Loeffler mentioned using a short course of prophylactic antibiotics postoperatively helped to rapidly resolve swelling [12].

Reported complications of TTS are tissue loss, flexion deformities, and amputation [13]. Postoperative complications include rotation of the soft tissue around the tip of the toe, causing the nail to shift in a medial direction [14]. Use of a single tacking stitch managed to resolve this and has been considered in the surgical management of TTS [14].

6. Discussion

Characteristically, in toe tourniquet syndrome, the hair is wrapped very tightly around a single toe or multiple toes of an infant [4,11]. Due to the highly constrictive nature of the hair, the lymphatic drainage from the distal toe is impeded, causing it to swell up resulting in oedema and redness of the toe. Furthermore, rising compression causes the venous return to halt and again further oedema develops. Finally arterial perfusion is impaired and causes ischemia which if not corrected can lead to tissue necrosis and eventually auto-amputation of that digit [8,11,13]. If TTS is identified and corrected early enough, there is a high chance of no long term damage occurring [1]. Early recognition is therefore a key issue in managing this syndrome.

If left untreated the condition, will most likely result in loss of function if not auto-amputation of the involved appendage and for this reason that a high index of clinical suspicion should be applied in order to avoid significant morbidity for the patient. The appendages described to have been involved include fingers, toes, penis, labia minora, clitoris and neck. However the great majority of cases described involve the toes, most often the 2nd and 3rd.

Differential diagnosis includes inflammation from infection, allergy, dermatitis, insect bite, trauma or idiopathic oedema or sexual abuse if the genitals are affected. The cause of such a case can be problematic to the diagnostician as they consider the involvement of child abuse [15]. There have been examples of cases in which the parents of the patient have admitted to inducing the tourniquet themselves as punishment [16]. Also written by Haddad are further cases of intentional harm involving cultural belief. Some cultures will wrap the hair purposefully onto the newborn's digits as they believe it will ward off evil spirits [16]. Klusmann and Lenard [17] have argued that cases involving abuse of the infant will have noticeable features such as a lack of explanation by the parents, when multiple digits or multiple strands are involved and the presence of knots within the hair or thread.

7. Acknowledgements

Acknowledgement to Mr. Umar Iqbal for editing the pictures for this article.

REFERENCES

- [1] J. D. Barton, G. M. Sloan, L. S. Nichter and D. J. Reinish, "Hair-Thread Tourniquet Syndrome," *Pediatrics*, Vol. 82, No. 6, 1988, pp. 925-928.
- [2] R. R. Miller, W. E. Baker and G. H. Brandeis, "Hair-Thread Tourniquet Syndrome in a Cognitively Impaired Nursing Home Resident," *Advances in Skin & Wound Care*, Vol. 17, No. 7, 2004, pp. 351-352. [doi:10.1097/00129334-200409000-00014](https://doi.org/10.1097/00129334-200409000-00014)
- [3] R. S. Strahlman, "Toe Tourniquet Syndrome in Association with Maternal Hair Loss," *Pediatrics*, Vol. 11, 2002, pp. 685-687.
- [4] S. Mackey, S. Hettiaratchy and J. Dickinson, "Hair-Tourniquet Syndrome—Multiple Toes and Bilaterality," *European Journal of Emergency Medicine*, Vol. 12, No. 4, 2005, pp. 191-192. [doi:10.1097/00063110-200508000-00009](https://doi.org/10.1097/00063110-200508000-00009)
- [5] J. J. Alpert, R. Filler and H. H. Glaser, "Strangulation of an Appendage by Hair Wrapping," *New England Journal of Medicine*, Vol. 273, 1965, pp. 866-867. [doi:10.1056/NEJM196510142731608](https://doi.org/10.1056/NEJM196510142731608)
- [6] J. H. Kuo, L. M. Smith and C. D. Berkowitz, "A Hair Tourniquet Resulting in Strangulation and Amputation of the Clitoris," *Obstetrics and Gynecology*, Vol. 99, No. 5, 2002, pp. 939-941. [doi:10.1016/S0029-7844\(01\)01753-7](https://doi.org/10.1016/S0029-7844(01)01753-7)
- [7] I. Claudet, N. Pasian, C. Debuissou, S. Salanne and H. Rekhroukh, "Tourniquet Syndrome: Interest of a Systematic Analysis of Families' Social Conditions to Detect Neglect Situations," *Child Abuse and Neglect*, Vol. 33, No. 9, 2008, pp. 569-572. [doi:10.1016/j.chiabu.2009.03.003](https://doi.org/10.1016/j.chiabu.2009.03.003)
- [8] S. Garcia-Mata and A. Hidalgo-Overjero, "Hair Tourniquet Syndrome of the Toe: Report of 2 New Cases," *Journal of Pediatric Orthopaedics*, Vol. 29, No. 8, 2009, pp. 860-864. [doi:10.1097/BPO.0b013e3181b7ff14](https://doi.org/10.1097/BPO.0b013e3181b7ff14)
- [9] D. D. Douglas, "Dissolving Hair Wrapped around an Infant's Digit," *Journal of Pediatrics*, Vol. 91, 2003, 1977, p. 162.
- [10] F. Serour and A. Gorenstein, "Treatment of the Toe Tourniquet Syndrome in Infants," *Pediatric Surgery International*, Vol. 19, No. 8, 2003, pp. 598-600. [doi:10.1007/s00383-003-1034-1](https://doi.org/10.1007/s00383-003-1034-1)
- [11] S. T. Sudhan, S. Gupta and C. Plutarco, "Toe-Tourniquet Syndrome—Accidental or Intentional?" *European Journal of Pediatrics*, Vol. 159, No. 11, 2000, pp. 866-874. [doi:10.1007/PL00008357](https://doi.org/10.1007/PL00008357)
- [12] R. A. Haene and M. Loeffler, "Hair Tourniquet Syndrome in an Infant," *The Journal of Bone & Joint Surgery*, Vol. 89-B, 2007, pp. 244-245.
- [13] R. Y. Liow, P. Budny and P. J. Regan, "Hair Thread Tourniquet Syndrome," *Journal of Accident & Emergency Medicine*, Vol. 13, No. 2, 1996, pp. 138-139. [doi:10.1136/emj.13.2.138](https://doi.org/10.1136/emj.13.2.138)
- [14] J. W. Mack, R. M. Takamoto, F. R. Jones, *et al.*, "Toe Tourniquet Syndrome," *The Western Journal of Medicine*, Vol. 125, 1976, pp. 335-336.
- [15] P. Lohana, G. N. Vashishta and N. Price, "Toe-Tourniquet Syndrome: A Diagnostic Dilemma!" *Annals of The Royal College of Surgeons of England*, Vol. 88, No. 4, 2006, pp. 6-8.
- [16] F. S. Haddad, "Penile Strangulation by Human Hair. Report of Three Cases and Review of the Literature," *Urologia Internationalis*, Vol. 37, No. 6, 1982, pp. 375-388.
- [17] A. Klusmann and H.-G. Lenard, "Tourniquet Syndrome—Accident or Abuse?" *European Journal of Pediatrics*, Vol. 163, 2004, pp. 495-498.