

Case Report

Hair Tourniquet Syndrome in a Young Infant

WCY WAN, MM YAU, DCY LAU, KF HUEN, LTW CHAN

Abstract Hair tourniquet syndrome is rare, referring to a condition when hair, thread or fiber wraps tightly around an infant's appendage causing a tourniquet effect, carrying a potential risk of permanent tissue damage or loss of appendage secondary to the strangulation. We report a young infant having hair tourniquet syndrome at the toe, with complete removal of the offending hair resulting in full recovery, however, no explanation for the strangulation identified. Possibility of child abuse and neglect has to be seriously considered for most if not all cases.

Key words Hair; Infant; Toe; Tourniquet

Case Report

A 73-day-old girl, enjoying good past health, presented to the accident and emergency department with a bluish swollen right fourth toe strangulated by a strand of hair at the metapalangeal joint. When the father changed clothes for his daughter, her toe was found to be swollen and the girl remained otherwise asymptomatic, without being irritable or having excessive crying. Failed to unloop the hair strand at the daughter's toe, the father sent the daughter to the hospital. On physical examination, the toe was cyanosed with poor circulation, devoid of tissue gangrene.

The infant was diagnosed to have hair tourniquet syndrome, removing the hair thread completely with the forceps and scissors by the orthopaedic surgeon in ward, resulting in full restoration of circulation and resolution of oedema of the toe. Figures 1 and 2 illustrate the constriction ring, after the removal of the hair thread, at the level of middle phalange of the slightly swollen right fourth toe. As non-accidental injury was suspected, the paediatrician was consulted for expert opinion. Full physical examination of this normal-sized infant was completely normal, without abnormal bruises, swellings or fractures. The head circumference, body length and weight were at the 50th, 75th, and 50th centiles respectively. She had good hygiene and normal nutritional status. The family was living in a privately-owned housing estate. The father was self-employed, the breadwinner of the household, working at home with stable income; the mother was a housewife taking care of three children, namely, the patient together with two elder siblings aged three, and one year old. Although the mother had history of depression, her mental state and emotion was currently stable, without concurrent psychiatric drug usage or excessive hair loss. No explanation for the strangulation was offered by the parents, and yet evidence of child abuse was lacking. The patient was discharged home on the next day after the parents were educated about the importance of regular examination of digits and genitals for hair entanglement.

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Figure 1 Constriction ring at right fourth toe with mild soft tissue swelling around.



Figure 2 Constriction ring at another angle.

Discussion

Hair or hair-thread tourniquet syndrome is the term coined to describe the condition when hair, thread or fiber wraps tightly around an infant's appendage causing a tourniquet effect.¹ 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.² 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. Tourniquet syndrome occurs infrequently, and the incidence is not known.³ Claudet et al⁴ 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 al⁵ 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. The apparent rare occurrence of the syndrome is probably underestimated as the patients may have sought medical attention from various medical disciplines: paediatricians, general practitioners, orthopaedic surgeons and doctors from the accident and emergency department. Our 73-day-old patient was firstly brought to the accident and emergency department, then referred to the orthopaedic surgeon and was finally under the care of us, paediatric team.

For tourniquet syndrome, toes and fingers are the most common sites of involvement while strangulations of genitals, notably penis and clitoris, or labial majora, have also been reported.^{4,5} Claudet found that majority of the tourniquet syndrome cases (95%) involved toes while the remaining common sites were penis and labial majora.⁴ 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.⁵ Moreover, they found that the third toe and middle finger were at greatest risk, followed by the index finger and second toe. Instead, our patient had strangulation at right fourth toe. The disparity in epidemiology of body parts involvement could be explained by the difference in data collection methods: Claudet revealed a single-centered experience while Barton's study represented a collection of reported cases in the literature.

Hair, the culprit in our case, was the chief offending agent concluded in both Claudet's study, noting that hair responsible for 95% of all cases;⁴ 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.⁵ 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.⁶ 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. 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,¹ during which the infant wallowed in the mother's hair or in

the fabric of sleeping bags, socks or mittens. Garments, having undergone repeated washing, are believed to cause strangulation of the digits easier as the threads have been frayed out. In our case, conversely, the mother had no history of excessive hair dropping, nor did our patient wear any socks. No explanation for the strangulation was identified.

Early recognition of diagnosing hair tourniquet syndrome is deemed necessary as strangulation of appendages can be a surgical emergency in which permanent tissue damage or loss of appendage can occur.⁷ Unfortunately, a delay of three or four days in diagnosis is common because irritability may be the only presenting symptom in a young infant who usually presents with excessive crying or redness of the extremity.^{1,8} 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.⁹ 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. High index of suspicion of hair tourniquet syndrome therefore has to be taken. Fortunately, the insult was diagnosed and treated early in our patient, without any permanent adverse consequences.

Conservative measures, removing the tourniquet, such as hair removal with fine forceps and scissors under magnification, or soaking the body part in depilatory cream to weaken the fibers, have been the standard of management for hair tourniquet syndrome.^{5,10} Nevertheless, conservative cuts using scalpels or needles in the accident and emergency department may not release all the constricting fibers as the involved body part may be swollen, and hence some authors advocate urgent complete release in the operating theatre.³ Thanks to the orthopaedic surgeon, the whole hair thread strangulating our patient's toe was successfully removed by fine forceps and scissors at bedside.

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, 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 recognised 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".¹¹ In fact, the earliest reports of tourniquet syndrome as a form of child abuse could be dated back to 1832.² A case series by Klusmann et al ascribed all five tourniquet syndrome cases to some form of child abuse.⁷ Without doubt, adopting a sensible and sensitive approach in handling young children presenting with unusual conditions is a must. Apart from taking a thorough history, performing a full physical examination, reviewing previous medical and social records, and liaising with relevant health professionals, different child care practices should be acknowledged, especially in a multicultural society, for instance, some tribes would wrap the penis to prevent nocturnal enuresis.¹¹ Next, evidence of other abuse, other unusual events, and possible abuse of siblings should be looked for. Although Claudet found only one child abuse case among his collection of cases, 67% of the patients with hair tourniquet syndrome were already followed by child protective services with extreme poverty or living in poor housing with insufficient hygiene, signifying a possible association between hair tourniquet syndrome and poverty and poor hygiene.⁴ Possible indicators of child abuse included genital strangulation, multiple and distant locations (e.g. toes and genitals), multiple and/or separated knots, as well as constrictive agents inconsistent with a safe environment for the child.^{1,4,5} Despite data correlating non-accidental injury with tourniquet syndrome in the literatures are deficient, child abuse has to be seriously considered for most if not all tourniquet cases. After all, reassessing the possibility of deliberate harm in every case is easier said than done as incorrect accusations of abuse may cause as much harm as failure to recognise.

As for all disease entities, prevention is always better than treatment and of the essence hair tourniquet syndrome can be preventable. First of all, postpartum mothers, particularly those with long hair, should be counseled about this rare condition and the possibility of excessive hair loss after delivery. Secondly, stringent measures of checking the toes and fingers by turning any covered clothing inside out after bathing or swimming before dressing the baby was suggested by Strahlman.¹ Needless to say, mothers should seek medical attention immediately if the first sign of entrapment or circulation compromise is noticed or suspected.

Conclusion

We report a young infant having hair tourniquet syndrome at the toe, with complete removal of the offending hair resulting in full recovery, however, no explanation for the strangulation identified. Early recognition facilitates effective treatment and reduces the potential risk of permanent tissue damage or loss of appendage secondary to the strangulation. Leave alone most reported hair tourniquet syndrome cases involving toes were accidental, a likely association was found between tourniquet cases and a lack of hygiene, extreme poverty and poor housing environment. Lastly the possibility of child abuse should be considered if injuries are involving the genitals, multiple and at distant locations, presence of multiple and/or separated knots, and suspicious home or social circumstances for the child.

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