

Hair tourniquet syndrome, an unusual presentation: A case report and review of literature

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ABSTRACT

Introduction: Hair tourniquet syndrome is rare presentations that are not often encountered in orthopedic OPD's. The syndrome was recognized long back but there have only been a few published reports in regard to the sequel it follows. The usual case presentation was an infant that had been crying in an inconsolable manner and upon careful examination was found to have a digit or a body part entangled by a hair or a textile thread causing a tourniquet like effect but our case was unique as it was an incidental finding. **Case Report:** This case reports a 5 months old healthy female child that was accidentally found to have a hair wrapped around her left fourth toe on her regular routine vaccination visit. **Conclusion:** This presentation is quite different than the presentation usually encountered or those presented previously in literature. The diagnosis of the syndrome requires a high index of suspicion, it may present a diagnostic dilemma therefore through awareness of the condition is important.

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INTRODUCTION

Hair thread tourniquet syndrome is an uncommon occurrence of events with an incidence reported to be around 0.02% [1]. It is seen when a hair strand or alternatively a textile thread may be the cause of strangulation around an appendage. The first reports of such a pathology were reported around 18th century in a 4-week old infant although it was recognized much earlier around in the 17th century [2]. Body parts such as finger, toe, glans penis, and vulva may be involved. Depending upon degree of strangulation it may lead to ischemic necrosis further being complicated by gangrene and amputation. The importance lies in early recognition and appropriate intervention before the onset of complications of permanent tissue necrosis.

We report a case of 5-month-old baby girl with a hair tourniquet syndrome affecting the fourth toe, there was no report of any symptoms from the parent's or child side and it was an incidental finding during routine vaccination.

CASE REPORT

A 5-month-old baby girl was incidentally found to have a swollen and erythematous left fourth toe upon routine vaccination and was referred to ER for appropriate management. She was diagnosed to have a hair tourniquet strangulating the fourth toe which was removed physically (cut by a scissors). Despite removal of the hair strand the swelling continued, she was re-evaluated after a week and diagnosed as superficial skin infection being started on antibiotics (oral and local) for 10 days. One month post initial presentation the patient was seen in our outpatient department due to persistent swelling Figure 1(A and B) where the diagnosis of hair tourniquet was revisited and patient was taken for formal exploration under GA. After proper skin preparation with betadine and drapping, skin incision was given over the dorsal aspect of the fourth toe at the site of skin dimpling. After superficial dissection, a strand of hair was found to be encircling the fourth toe circumferentially. It was removed completely and skin was sutured using monocryl 3/0. The offending agent was the hair strand that had cheese wired through the skin and was strangulating the toe from within (Figures 2, 3). The child was discharged in stable condition, at follow-up in 1 month time the toe had returned to its complete function and the redness along with swelling had subsided Figure 4(A and B).

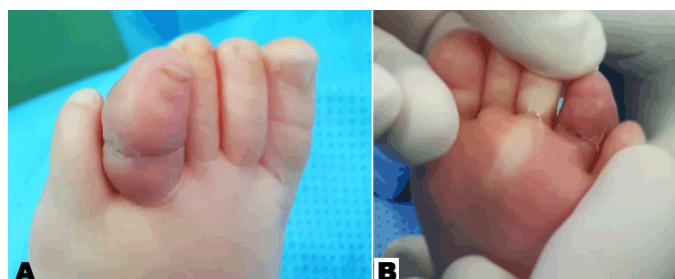


Figure 1(A and B): Pre-Operative images of the left foot with the tourniquet band being shown completely encircling both the dorsal and plantar surface of the fourth toe.



Figure 2: Intra-operative image showing the deep band.



Figure 3: Cheese-wiring of the hair being clearly demonstrated as the cause of tourniquet is embedded deep in the subcutaneous tissues.



Figure 4(A and B): Post-operative follow up in one month's time shows completely healed wounds.

DISCUSSION

The term hair tourniquet syndrome was initially coined by Barton in 1988 [3], although it was recognized earlier. There are almost 200 reported cases of hair tourniquet syndrome to date [2] with most involving the genitalia followed by toes, fingers and other sites [3]. Up to now 2 large case series have been published, the first one was by Barton et al (1988) [3], where he reported 60 cases from his center, another large series of case reports was reported by Golshevsky et al (2005) [4] where he looked upon 90 cases from different centers. A meta-analysis of major case reports was done by Arman saad et al (2009) [5] where upon he looked at 259 involved appendages. The frequency of appendage involvement in descending order were penile (44.2%), toe (40.4%), finger(8.57%), other sites(female genitalia, uvula, neck)(6.83%). The third toe was reported to be most frequently involved followed by fourth toe/second toe, little toe. [5, 6]

The term toe tourniquet syndrome was coined by Qunin [7] in 1971 to specifically describe conditions in which strand of hair or thread may wrap around a digit distally causing ischemic damage to the under lying

digit. Although hair tourniquet syndrome is classically described in younger infants, there has been reports of its occurrence in elderly population (above 6th decade), in a case report a 9 year old as reported by okeke et al (2009) [2, 8, 9]. The typical age of occurrence is less than 1 year with a mean of 5.5+/- 4 months. Finger involvement is more common in first few weeks of life followed by toe involvement in slightly older infants [5]. Penile involvement is seen more in patients above 2 years of age [3, 5].

There has been a lot of discussions regarding the pathophysiology of the syndrome but to date a consensual explanation is still pending, there are various theories circulating in terms of etiology of the disorder. The initial one was put forward by Quinin himself where he argued that the entity is caused by usage of certain type of clothing material (leotard type) in combination with plantar reflexes of child [5, 7]. It was seen that for toes and genitalia the offending agent was mostly hair strands whereas in fingers it is mostly caused by mittens. Alpert et al in 1965 in his report suggested that the high tensile strength of hair makes it an effective tourniquet as the hairs are stretchable when wet and recoil when dry [5, 10]. This was further re-affirmed by Strahlam et al [11] who in 2003 pointed out the association between the natural postpartum telogen effluvium and the hair tourniquet syndrome [6, 12]. Although majority of the cases are non-accidental but Kulsman et al in 2004 [6, 13] reported four cases of child abuse presenting as hair tourniquet syndrome. They argued the presence of multiple knots within the hair, multiple digits being affected and a lack of explanation. It is however still debatable so as to how many such injuries can be accounted as non-accidental but these factors can be used as an indicator for further investigations. There is no sex predilection for the toe tourniquet syndrome although it is unknown to why fourth toe is most commonly affected.

The clinical presentation is a subtle one, it may range from swelling and redness to excessive crying and irritability [5-7]. The most common presentation was the latter one. The period to presentation is variable but in most reports the average period to presentation was 48 hours following excessive crying and irritability [14]. Our case was unique in this aspect where findings were incidental altogether and parents did not report any symptoms of irritability. The swelling was initially noticed by health care workers when the child went for her routine vaccination which has not be seen previously in literature therefore we stress the need of high index of suspicion. The offending fibers may not be always visualized under direct vision occasionally the hair may cheese wire through the soft skin where it may be buried and obscured by re-epithelization of skin but the tourniquet effect still continue causing lymphatic obstruction, tissue-edema leading to venous congestion and ultimately affecting arterial perfusion, this starts a vicious cycle of events where more tissues tend to continue to swell

causing the tourniquet to be tighter and tighter [5, 15]. Delayed diagnosis may lead to gangrene and amputation in certain cases. The sequence of events may occur insidiously over a couple of months or rapidly progress therefore timely intervention is of prime importance. Initial management consists of examining the child in properly lit area, ascertain the nature of constricting ring (hair/thread, multiple threads or multiple toes involved). Once the diagnosis is established complete removal of the offending agent is necessary. There are multiple reports by Alruwaili et al [15] where they have used a depilatory agent to remove the hairs to release the tourniquet but the efficacy of such agent is still questionable, due to their non-availability in many ER and also in severely edematous tissues the complete removal of hairs cannot be ascertained. This point was elaborated by Kerry et al [16] who suggested surgical release in such cases however there are many such cases in which complete release was not achieved in the first attempt and surgical revision was necessary. Our case is a good example where an initial attempt for removal was done in another center, later on the patient was treated as secondary infection although the pathology was incompletely removed due to cheese wiring of the hairs under the skin and re-epithelization. Surgical release may be done using single lateral incision as advocated by Karry and chapman et al Or by using double incision where contralateral incision is done down to the bone to completely remove the offending agent. Such approach has been advocated by Mata et al [2, 17]. Our case was dealt using dual approach, intra-op the affecting agent (hair) was clearly seen embedded under the skin causing the tourniquet effect subcutaneously. Post-decompression the prognosis is excellent ascertaining the fact that in infants the propensity of soft tissues to heal is far better than adults never the less timely diagnosis should be made to avoid complications such as flexion deformity, auto-amputation and gangrene as a result of prolonged hair tourniquet syndrome.

CONCLUSION

Hair tourniquet syndrome is neither that rare nor new but still it remains poorly recognized it may present as a diagnostic dilemma especially when the hair might have been buried under the skin, however it must be considered as a differential in evaluating an excessively irritable infant or if child presents with swollen digit or when there is circumferential depression proximally over the digit. Mothers may be counseled regards the post-partum telogen effluvium. It is also advocated to advise the mothers that certain types of clothes should be laundered inside out to avoid harboring of threads or human hairs. A vigilant examination and timely intervention cannot be stressed enough to avoid unwanted complications.

ABBREVIATIONS

OPD: out-patient department, ER: emergency room,
GA: general anesthesia

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Author Contributions

Sadaf Mohammed Basheer – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Ali Redha Gerashi – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Mohammed Abdul Basith – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor of Submission

The corresponding author is the guarantor of submission.

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Consent Statement

Written informed consent was obtained from the patient for publication of this case report.

Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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