Hair-thread strangulation syndrome in childhood: a systematic review

Daniela Djokic a, Gregorio P. Milani b, Sebastiano A. G. Lava c, Gianluca Gualco d, Teresa Corigliano d, Mario G. Bianchetti d, Camilla Lavagno g

Summary

INTRODUCTION: Hair-thread strangulation syndrome describes the constriction of a body part by a tightly wound hair or thread. This research aims to review the literature about this entity.

METHODS: A systematic review was performed to characterise hair-thread strangulation syndrome in subjects aged ≤16 years. This pre-registered review (PROSPERO ID: CRD42022363996) followed the PRISMA methodology.

RESULTS: Subjects with digital strangulation were significantly younger (median = 4.0 [interquartile range: 2.0–6.1] months; n = 143) than females with genital strangulation (9.0 [6.8–11] years; n = 36), males with genital strangulation (5.1 [1.9–6.0] years; n = 36), and subjects with nondigital and non-genital strangulation (24 [13–48] months; n = 11). Digital strangulation was followed by an amputation in five (3.5%) and a reconstructive surgical intervention in seven (4.9%) cases. Sequelae occurred in four (11%) cases after female genital strangulation: clitoris autoamputation (n = 2) and surgical removal of a necrotic labium minus (n = 2). Severe complications were observed in 14 (39%) cases with male genital strangulation: urethral fistula (n = 7), urethral transection (n = 2), and partial penile autoamputation (n = 5). A partial urological autoamputation was observed in one case (9.0%) with non-digital and non-genital strangulation.

CONCLUSIONS: Early recognition and management are crucial to avoid sequelae or long-term care in hair-thread strangulation syndrome.

Introduction

Hair-thread strangulation syndrome describes the acute constriction of a body part, such as digits or external genitalia, by a tightly wound hair or thread in a young infant, a child, or an adolescent [1]. Narrow constriction reduces lymphatic and venous drainage, resulting in pain, swelling, oedema, and, if recognised late, ischemia [1]. This condition was first observed in the 19th century in a 4-week-old English boy with a painful penis swelling induced by a circumferential hair proximal to the swelling [2]. Hair-thread strangulation syndrome is either not mentioned or only briefly discussed in currently available paediatric textbooks. Its symptoms and presentation may be misdiagnosed, leading to delayed treatment and potentially adverse outcomes. A comprehensive literature review on this condition could help raise awareness among healthcare providers, ultimately improving diagnostic accuracy and patient outcomes. Therefore, we conducted a systematic review on this paediatric syndrome.

Methods

Search strategy

This literature review did not require Institutional Review Board approval. It was pre-registered at the International Prospective Register of Systematic Reviews (PROSPERO ID: CRD42022363996) and followed the 2020 edition [3] of the Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines (PRISMA) according to the Joanna-Briggs Manual. The search was undertaken in the National Library of Medicine, Excerpta Medica, and Web of Science databases without restriction. The search strategy combined the following terms: “hair strangulation syndrome” OR “hair tourniquet syndrome” OR “hair-thread strangulation syndrome” OR “hair-thread tourniquet syndrome” OR “strangulation syndrome” OR “tourniquet syndrome”.

Articles available in Google Scholar or already known to the authors were also considered. The search was undertaken in February 2022 and repeated before submission (November 2022).

Following a first round of selection based on title and abstract, the full text of the selected articles was evaluated for eligibility.
Eligibility criteria and data extraction

Eligible articles were original reports and letters published in peer-review journals describing individual subjects aged ≤16 years with hair-thread strangulation affecting the digits, the external female genitalia, the penis, or other appendages. Cases with rubber band syndrome around the wrist were excluded [4]. Cases published only as abstracts and case series not detailing information on individual cases were also excluded.

The extracted data included demographics, the anatomic location, the source of strangulation (hair, threads, or elastic bands), the possible deliberate strangulation (by a caregiver or self-inflicted), the necessity of a reconstructive surgical procedure, and the outcome (development of long-term sequelae).

Two investigators independently (and in duplicate) performed the literature search, the selection of reports retained for analysis, and the data extraction in an unblinded manner with the support of a senior investigator. Any disagreements were discussed. One investigator entered the data into a pilot-tested database, and another verified them for accuracy.

Analysis

Pairwise deletion was used for missing data. Discrete variables are presented as counts and compared using Fisher’s exact test. Numerical variables are presented as medians and interquartile ranges or as box-and-whisker plots and compared using Kruskal–Wallis and Tukey’s tests [5, 6]. Two-sided P-values of <0.05 were considered statistically significant. Data were analysed using GraphPad Prism (version 9.5.1).

Results

Search output

The literature search identified 231 potentially relevant articles (figure 1). After removing irrelevant articles, 168 full-text articles were reviewed for eligibility. One hundred fifty-two articles [2, 7–157] describing individual cases of hair-thread strangulation syndrome published between 1951 and 2022 in English (n = 147), German (n = 2), French (n = 1), Spanish (n = 1), and Dutch (n = 1) were retained for the final analysis.

Fifty-eight articles reported cases from America, 45 from Asia, 39 from Europe, six from Oceania, and four from Africa. The articles provided information on 226 cases: 143 cases of digital strangulation [2, 7–90], 36 of female genital strangulation [22, 36, 49, 86, 91–119], 36 of male genital strangulation [7, 12, 24, 120–147], and 11 cases of non-digital and non-genital strangulation [148–157]. Characteristics of the individual cases are provided in the supplementary material (supplementary tables 1–4).

Clinical data

General data

Subjects with digital hair-thread strangulation syndrome (females: 50%; males: 50%) were significantly (p < 0.01) younger (4.0 [2.0–6.1] months) than females with genital strangulation (9.0 [6.8–11] years), males with genital strangulation (5.1 [1.9–8.0] years), and subjects (females: 58%; males: 42%) with non-digital and non-genital strangulation (24 [13–48] months; figure 2). Females with genital strangulation, males with genital strangulation, and subjects with non-digital and non-genital strangulation did not differ significantly in age.

Twenty-nine (13%) of the 226 cases were explicitly reported as not induced by hairs but by threads arising from footed pyjamas, mittens, socks, underwear, and a nylon or elastic band. Three (1.3%) further cases were caused by hairs and threads.

The prevalence of cases not associated only with hairs was significantly higher (p = 0.0003) for digital strangulation (n = 29; 20%) than for the other forms of strangulation syndrome (n = 3; 3.6%).

A non-accidental cause of strangulation was suspected in 21 cases [12, 17, 36, 132, 133, 143]. Voluntary penile constriction was induced to manage enuresis in three cases [132, 133, 143].

Digital strangulation syndrome

Patients with digital hair-thread strangulation syndrome presented with irritability, crying, digital swelling, discoloration, and pain. About four fifths of the 143 cases of digital strangulation syndrome were observed on the toes (table 1). The female-to-male ratio did not differ significantly (p = 0.336) between infants with toe compared to finger strangulation syndrome. Cases with toe strangulation were slightly but significantly older than those with finger strangulation by 1.5 months on average (p = 0.034).

A detailed description of the affected digits was available for 136 (95%) of the 143 cases. Finger strangulation al-
ways affected only one finger. In contrast, toe strangulation affected at least two toes in about a third of cases. The little finger, the big toe, and the little toe were relatively rarely affected. Digital strangulation was induced by threads in eight (38%) of 21 cases with finger strangulation and in 12 (10%) of 115 cases with toe strangulation ($p = 0.0037$).

An amputation was performed in five (3.5%) cases: left thumb ($n = 1$), left middle toe ($n = 1$), right big toe ($n = 1$), right middle toe ($n = 1$), and right little toe ($n = 1$) [12, 26, 30, 53, 88]. A reconstructive surgical intervention was necessary in seven (4.9%) cases [12, 28, 29, 41, 70, 74, 81]. Removal of hair thread either mechanically or by applying a depilatory agent was followed by a full recovery in the remaining 131 (92%) cases.

**Female genital strangulation syndrome**

Hair-thread strangulation syndrome of the female external genital was reported in 36 cases, who presented with genital pain (sometimes associated with wide-base gait or painful micturition), vulvar swelling, or both genital pain and vulvar swelling. The strangulation was localised as follows (two structures were concurrently affected in two cases): clitoris ($n = 19$), labium minus ($n = 16$; bilateral in one case), labium majus ($n = 2$), and mons pubis ($n = 1$). Sequelae were observed in four (11%) cases: total clitoris autoamputation in a 14-year-old adolescent, partial clitoris autoamputation in a 5-year-old girl, and surgical removal of a necrotic labium minus in a 6-year-old and a 10-year-old girl [95, 97, 105, 119]. The remaining 32 cases recovered uneventfully.

**Male genital strangulation syndrome**

Hair-thread strangulation syndrome of the male external genital was reported in 36 cases. Young infants presented with irritability and crying, often associated with swelling; older children presented with pain and swelling.

Table 1:

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*p = 0.343 and **p = 0.034 versus toe strangulation.

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**Figure 2:** Age at presentation in paediatric patients with the various forms of hair-thread strangulation syndrome. The results are presented as box and whisker plots. Please note that the vertical axis is on a logarithmic scale. The age at presentation was significantly lower in children with digital strangulation than in children with strangulation of genitalia ($p < 0.005$) or other appendages ($p < 0.01$).
Severe complications were observed in 14 (39%) cases [121, 123, 127, 128, 130, 134–136, 141, 143, 145–147]: urethral fistula (n = 7), urethral transection (n = 2), and partial penile autoamputation (n = 5).

**Non-digital and non-genital strangulation syndrome**

A non-digital and non-genital strangulation syndrome was noted in 11 cases (female-to-male ratio = 1.3): uvula (n = 5), neck (n = 3), circumvallate tongue papilla (n = 2), and tooth (n = 1). A partial uvular autoamputation was observed in one case [157].

**Discussion**

This literature review addressed the entire spectrum of conditions that may be caused by hair-thread strangulation in paediatric patients. While it found that they mainly affected the digits of young infants, they could also occur in older children. It also demonstrated that severe sequelae or the need for long-term care are possible (especially in cases with digital or penile strangulation). Furthermore, strangulation is sometimes non-accidental (e.g. intentional application to manage bedwetting). Finally, some cases were not caused by hairs but by threads or an elastic band.

Infants with hair strangulation often presented with a history of acute excessive crying [158, 159]. While the cause of acute and unexplained infantile crying is often benign, a careful history and physical examination remain the cornerstone of the evaluation. However, many cases present with local symptoms and signs such as pain, swelling, or discolouration. Detecting the encircling hair (which might be more challenging with light-coloured hairs) or thread is critical for diagnosis.

Because of the risk of ischemic damage, rapid management of hair-thread strangulation syndrome is crucial [160, 161]. Our review did not specifically address this issue. After taking a history and performing an examination to make the diagnosis, the first management step is currently appropriate analgesia [162]. Chemical depilatory agents, which break hairs (or threads) and subsequently lead to a weakening of the fibres and eventual dissolution, are currently the first-line treatment choice because they are generally painless and noninvasive [160, 161]. Mechanical release should be performed if strangulation release is not achieved after two attempts with a depilatory agent. Cases with strangulation near mucous membranes or with signs of ischemia should be treated with primary mechanical release [160, 161].

In infants, strangulation was caused mainly by maternal hair related to increased hair loss after birth [163]. It is assumed that hair may fall unnoticed into the bath water during routine care or diaper changing. Our review had both weaknesses and strengths. Its main weakness was the low number of identified cases. The second weakness was its inability to address potential risk factors, such as socioeconomic status or ethnicity. Its most relevant strength was that it covered the entire spectrum of conditions that may be caused by hair-thread strangulation in paediatric patients. For example, two recent reviews focused explicitly on the hair-thread strangulation syndrome of the female genital tract or both the female and the male genital tracts [118, 164].

**Conclusions**

Early recognition and management are crucial to avoid sequelae or long-term care in hair-thread strangulation syndrome. Children and infants with unexpected acute onset of crying or local symptoms and signs such as pain, swelling, or discolouration require rapid medical evaluation. Healthcare professionals should be aware of this condition, with a particular focus on digital strangulation in infants and genital strangulation in children.

**Data sharing statement**

The data underlying this article can be shared upon reasonable request to the corresponding author.

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**Author contributions:** Drs. Milani, Bianchetti and Lavagno conceptualized and designed the review. Drs. Djokic, Milani, Lava and Lavagno acquired the data and performed the analysis. Drs. Djokic, Bianchetti and Lavagno drafted the initial manuscript. All authors reviewed and revised the manuscript and approved the final manuscript as submitted.

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**Potential competing interests**

All authors have completed and submitted the International Committee of Medical Journal Editors form for disclosure of potential conflicts of interest. No potential conflict of interest related to the content of this manuscript was disclosed.

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Appendix: Supplementary tables
### Supplementary table 1:

Characteristics of 143 individuals affected by digital hair-thread strangulation syndrome. The country of publication is identified using the International vehicle registration code.

<table>
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### Supplementary table 2:
Characteristics of 36 female subjects affected by genital hair-thread strangulation syndrome. The country of publication is identified using the International vehicle registration code.

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Supplementary table 3:
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Supplementary table 4:
Characteristics of 11 individuals affected by non-digital and non-genital hair-thread strangulation syndrome. The country of publication is identified using the International vehicle registration code.

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