

Hair-thread strangulation syndrome in childhood: a systematic review

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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 [1.9–8.0] years; n = 36), and subjects with non-digital 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 uvular 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 condi-

tion 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.

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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%).

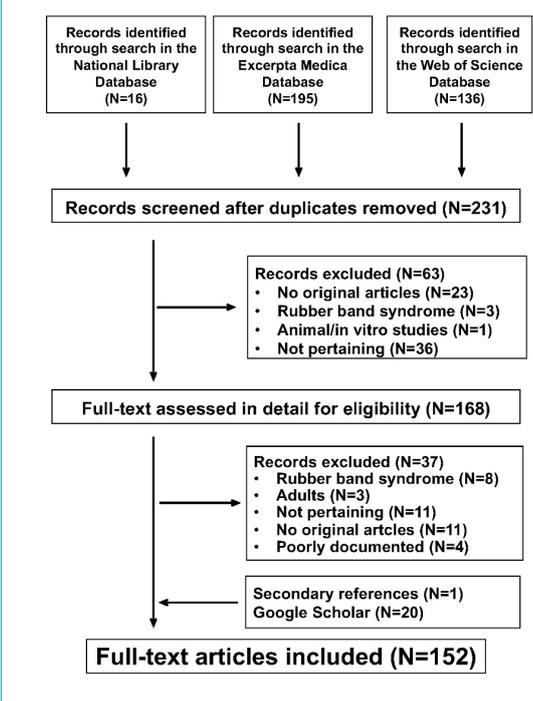
A non-accidental cause of strangulation was suspected in 21 cases [12, 17, 36, 132, 133, 143]. Voluntary penile constriction was induced to manage enuresis in three cases [132, 133, 143].

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-

Figure 1: Digital, female genital, male genital, and non-digital non-genital hair-thread strangulation syndrome in childhood. PRISMA flowchart of the literature search.

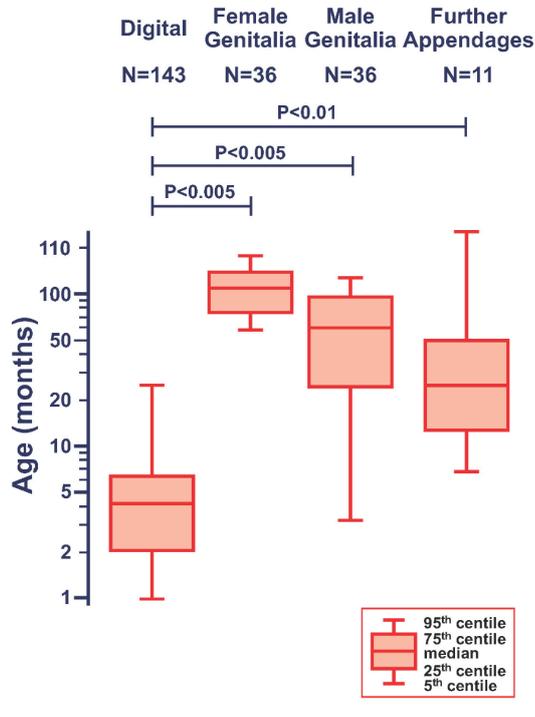


ways affected only one finger. In contrast, toe strangulation affected at least two toes in about a third of cases. The little finger, the big toe, and the little toe were relatively rarely

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.

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 ($p < 0.01$).



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: Fingers and toes with hair-thread strangulation syndrome in 136 paediatric cases.

		Total	Right	Left
Fingers				
Female:male ratio	0.6*			
Age	2.4** [2.0–3.1] months			
n		21	10	11
Thumb		7	3	4
Forefinger		3	1	2
Middle finger		6	4	2
Ring finger		5	2	3
Little finger		0	0	0
One finger		21	10	11
Two fingers		0	0	0
Three fingers		0	0	0
Toes				
Female:male ratio	1.1			
Age	4.0 [2.6–5.0] months			
n		115	67	48
Big toe		3	2	1
Long toe		28	19	9
Middle toe		82	46	36
Ring toe		45	24	21
Little toe		8	5	3
One toe		79	47	32
Two toes		21	11	10
Three toes		15	9	6

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

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

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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.

Case	First author	Year of publication	Country	Sex	Age (months)
1	Alpert JJ [7]	1965	USA	Male	2.0
2				Male	19
3	Curran JP [8]	1966	USA	Male	2.5
4	Chonwn [9]	1967	CDN		12
5	Hill RH [10]	1967	CDN		5.0
6	Hack M [11]	1972	IL	Female	0.8
7	Kerry RL [12]	1973	USA	Male	4.0
8				Male	1.8
9				Male	5.0
10				Female	2.3
11				Male	1.5
12				Female	1.8
13				Female	3.5
14	Mack JW [13]	1976	USA	Female	11
15				Male	5.0
16	Douglas DD [14]	1977	USA		10
17				Female	11
18	Miller PR [15]	1977	USA	Female	15
19	Arato A [16]	1978	H		6.0
20	Narkewicz RM [17]	1978	USA	Male	4.0
21				Male	3.0
22				Male	1.0
23	Mosely LH [18]	1979	NZ	Female	2.0
24	Bérard J [19]	1981	F	Male	6.0
25				Male	3.0
26				Male	3.0
27	Manfemaleredi SJ [20]	1981	USA	Female	7.0
28				Male	3.0
29	Barton DJ [1]	1988	USA	Male	5.0
30				Female	2.0
31				Female	4.0
32				Male	1.8
33				Male	144
34				Female	0.6
35	Collins AG [21]	1990	AUS	Female	14
36	Beasley SW [22]	1993	USA		2.5
37	Liow RY [23]	1996	UK	Female	18
38				Male	3.0
39	Vazquez Rueda F [24]	1996	E	Female	2.0
40				Male	4.0
41				Male	2.0
42				Female	5.0
43				Male	3.0
44				Male	7.0
45	Connors G [25]	1997	USA	Male	5.0
46	Lefemaleerink VJ [26]	1997	NL	Male	4.0
47				Male	4.0
48				Female	2.0
49				Male	0.8
50	Debo HF [27]	1999	USA	Male	2.0
51				Male	4.0
52	Sudhan ST [28]	2000	UK		5.0
53	Sunil TM [29]	2001	IND	Female	8.0
54	Wang M [30]	2001	USA		1.5
55	Corazza M [31]	2002	I	Female	72
56	Harris EJ [32]	2002		Female	5.0
57				Male	5.0
58	Smith AM [33]	2003	UK	Male	7.0
59	Strahman RS [34]	2003	USA	Male	3.0
60	Karalezli K [35]	2004	B	Male	3.0
61	Klusmann A [36]	2004	D		4.0

62					4.0
63					1.5
64					2.0
65	Mackey S [37]	2005	UK	Female	3.0
66	Gutiérrez-Gómez C [38]	2006	MEX	Female	2.0
67	Kurup HV [39]	2006	UK	Male	3.0
68	Lohana P [40]	2006	UK		1.5
69	Mat Saad AZ [41]	2006	IRL	Female	3.0
70	O'Quinn JC [42]	2006	USA	Female	2.3
71	von Känel OE [43]	2006	CH	Male	2.0
72	Anwar MU [44]	2007	UK	Female	4.0
73	Haene RA [45]	2007	UK		2.8
74	Kumaravel S [46]	2008	IND		24
75	Singh V [47]	2008	UK	Female	14
76	Al-Buainain H [48]	2009	KSA	Male	3.0
77	Cutrone M [49]	2009	I	Female	2.0
78				Female	2.0
79	Garcia-Mata S [50]	2009	USA	Female	18
80				Male	5.0
81	Ünlü C [51]	2009	NL	Male	5.0
82	Cevik Y [52]	2010	TR	Female	4.0
83	Heather J [53]	2011	NZ	Female	36
84	O'Gorman A [54]	2011	USA	Male	3.0
85	Onyeama CO [55]	2011	USA	Male	3.0
86	Sivathanan N [57]	2012	UK		
87					
88	Shankar R [56]	2012	IND	Female	1.0
89	Wan WCY [58]	2012	CHN	Female	2.0
90	Alvarez-Perez A [59]	2013	E	Male	8.0
91	Hickey BA [60]	2013	UK	Male	48
92	Au S [61]	2014	USA	Male	6.0
93	Bizzotto N [62]	2014	I	Female	2.0
94	Kamal NM [63]	2014	KSA	Female	6.0
95				Male	6.0
98	Baloch N [65]	2015	PK	Female	4.0
99	Bastug O [66]	2015	TR	Female	0.5
100	El-Kehdy J [67]	2015	RL	Female	6.0
101					
102	Hussin P [68]	2015	MAL	Female	3.0
103				Male	5.0
104	Kuiper JW [69]	2015	NL	Female	18
105	Pinkowsky GJ [70]	2015	USA	Female	84
106	Pye S [71]	2015	UK	Female	48
	Alruwaili N [64]	2016	KSA	Male	6.0
					0.5
107	van Meurs A [72]	2016	USA	Female	4.0
108	Dunphy L [73]	2017	UK	Male	2.0
109	Ojha S [74]	2017	IND	Female	12
110	Rahul SK [75]	2017	IND	Male	3.0
111	Tengattini V [76]	2017	I	Female	8.0
112	Uygur E [77]	2017	TR	Female	4.0
113				Female	6.0
114	Yadav R [78]	2017	IND	Male	2.0
115	Booth J [79]	2018	UK	Male	3.0
116					14
117	Ihara T [86]	2018	J	Female	1.3
118	Sebaratnam D [81]	2018	E	Female	24
119	Singh AP [82]	2018	IND	Male	1.5
120	Agherbi F [83]	2019	MA		
121	Aslantürk O [84]	2019	TR	Male	2.0
122				Male	5.6
123				Female	1.9
124				Female	3.5
125				Female	0.6

126				Male	4.3
127				Male	2.0
128				Female	4.0
129				Male	1.0
130				Male	13
131				Male	0.8
132				Male	3.9
133				Female	6.0
134				Female	2.8
135				Female	2.6
136				Female	9.1
137	Basheer SM [85]	2019	BRN	Female	5.0
138	Kesu Belani L [80]	2019	MAL	Female	2.0
139	Azizalrahman A [87]	2020	KSA	Female	3.0
140	McGrath RB [88]	2020	IRL	Male	12
141	Shafemaleiq MA [89]	2020	MAL		2.0
142	Sutcu M [90]	2020	TR	Female	2.0
143				Male	0.5

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.

Case	First author	Year of publication	Country	Age (years)
1	Chapman HL [91]	1968	CDN	9
2	Summers JL [93]	1973	USA	11
3	Webley JA [94]	1973	USA	7
4	Press S [92]	1980	USA	8
5	Beasley S.W [22]	1991	AUS	8
6	Rich MA [95]	1999	USA	5
7	Sylvestzrak MS [96]	2000	USA	5
8	Kuo JH [97]	2001	USA	14
9	Golshevsky J [100]	2004	AUS	14
10	Klusmann A [36]	2004	D	12
11	Bacon JL [99]	2005	USA	13
12	Osterhoudt KC [101]	2005	USA	8
13	Alverson B [102]	2007	USA	9
14	Serour F [98]	2007	IL	4
15	Cutrone M [49]	2009	I	0.5
16	Pomeranz M [103]	2009	IL	11
17	Stock C [104]	2012	USA	12
18	Bannier MA [105]	2013	NL	6
19	Dua A [106]	2013	USA	12
20	Lee JH [107]	2013	UK	11
22	Ozcakir E [108]	2014	TR	9
22	Panagidis A [109]	2015	GR	14
23				10
24	Parlak M [110]	2015	TR	6
25	Zimmerman LN [111]	2015	USA	5
26	Zhong N [112]	2017	USA	10
27	Ihara T [86]	2018	J	14
28	Roman E [113]	2018	AUS	12
29	Erikci VS [114]	2019	TR	8
30				7
31				9
32	Imam AS [115]	2019	USA	6
33	Diaz-Morales O [116]	2020	E	10
34	Minodier P [117]	2020	F	5
35	Chawla RK [119]	2021	USA	10
36	Adjei N [118]	2021	USA	11

Supplementary table 3:

Characteristics of 36 male subjects affected by genital hair-thread strangulation syndrome. The country of publication is identified using the International vehicle registration code.

Case	First author	Year of publication	Country	Age (months)
1	DeWeese JA [120]	1952	USA	22
2	Alpert JJ [7]	1965	USA	13
3	Farah R [121]	1973	USA	132
4	Kerry RL [12]	1973	USA	2
5				4
6				10
7	Haddad FS [122]	1982	USA	24
8				24
9	McClure WJ [123]	1985	USA	60
10	Sheinfeld J [124]	1985	USA	84
11	Mariani PJ [125]	1986	USA	60
12	Vazquez Rueda F [24]	1996	E	24
13				48
14	Pantuck AJ [126]	1997	USA	36
15	Bangroo AK [127]	2005	IND	120
16	Chaware SM [128]	2006	IND	84
17	Dar NR [129]	2007	PK	60
18	Hussain HM [130]	2008	AUS	72
19	Okeke LI [131]	2008	NGR	108
20	Basner A [132]	2012	D	2
21	Pahwa HS [133]	2013	IND	96
22	Jesus LE [134]	2014	BR	120
23	Dikshit VK [135]	2015	IND	120
24	Gazali ZA [136]	2015	IND	72
25	Zengin K [137]	2015	TR	96
26	Sekhon V [138]	2017	IND	84
27	Sheikh Andalibi MS [139]	2017	IR	54
28	Arshadi H [141]	2019	IR	108
29	Degheili JA [142]	2019	RL	48
30	Ozcift B [140]	2019	TR	48
31	Bouassida K [143]	2020	TN	108
32	Rawls WF [144]	2020	PK	24
33				17
34	Loloi J [147]	2021	USA	96
35	Patterson JW [145]	2022	USA	0.5
36	Rampersad B [146]	2022	TT	24

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.

Case	First author	Year of publication	Country	Sex	Age (months)
1	McNeal RM [148]	1987	USA	Male	13
2	Krishna S [149]	2003	USA		3
3	Chegwidden HJ [150]	2005	USA	Female	11
4	Schneider K [151]	2013	USA	Female	84
5	Flores JR [152]	2014	MEX		24
6					48
7	Alghamdi F [153]	2018	KSA	Female	48
8	Wong K [154]	2019	USA	Male	13
9	Chan LYF [156]	2020	HK	Male	21
10	Stansfield J [155]	2020	UK		9
11	Leighton J [157]	2021	UK	Female	36