

Cutaneous Pili Migrans in a 25-Year-Old Man: A Case Report and Mini-Review of the Literature

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Abstract

Background: Cutaneous pili migrans (CPM) is a skin condition in which the hair shaft or fragment penetrates the epidermis or middle dermis and gives rise to a similar eruption with Cutaneous lava migrans (CLM). It is an extremely rare condition with less than 50 cases being reported to date.

Methods: We report a 25-year-old Chinese man who complained of a 3-week history of slightly painful and slow-moving black, meandering, threadlike lesion on the left sole. And give a mini-review of the literature on CPM.

Results: The lesion started at the base of the fifth toe and extended towards the heel. On physical examination, a grey-black, 8-shaped, serpiginous, linear eruption could be seen on the sole of left foot. At the leading end of the lesion, a black line be seen through the surface of the skin. A black-brown stripe was clearly visible in careful inspection by dermatoscopy, with neither tunnels nor worms were observed. After a minimally invasive surgery, the black line was removed and demonstrated to a naked hair without follicle, leading to a diagnosis of CPM.

Conclusion: CPM is a hair inducing migrating skin eruption mimicking CLM. Careful inspection and dermatoscopic examination can contribute to the diagnosis. Removal of the causative hair shaft can lead to recovery. Therefore, dermatologists should pay attention to differentiate it with other conditions which manifested as creep eruption.

Keywords: Cutaneous pili migrans; Cutaneous lava migrans; Creeping eruption

Introduction

Creeping eruption is a cutaneous manifestation characterized clinically by a linear or serpiginous cutaneous track that is slightly raised, erythematous and mobile [1]. Before 1957 when Yaffee et al. [2] first described a case in which an embedded hair formed a creep eruption, this term was generally used to refer to the parasitic infection, hookworm-related CLM. As more similar cases were reported, it has been suggested that creeping eruption may have other causes than parasitic diseases and, it is necessary to differentiate creeping eruption and CLM [1]. Since 1975, more than 40 cases of hair inducing creeping eruption have been reported by using various terms such as ‘imbedded hair’ [2,3], ‘hair fragment in the skin’ [4], ‘burrowing hair’ [5], ‘bristle migrans’ [6], ‘creeping hair’ [7], ‘migrating hair’ [8] and ‘cutaneous pili migrans’ [9-20]. Some scholars believe that among the above terms, ‘cutaneous pili migrans’ can better reflect the etiology and clinical characteristics of the condition. Herein, we present such a rare case and review the published literature briefly.

Case Presentation

A 25-year-old otherwise well Chinese man who came back from America presented with a 3-week history of a slow-moving, black, thread-like eruption on the left sole in 2019. He stated that the lesion generally asymptomatic most of the time, but slight pain could be felt when the black line moved obviously. The history was negative for getting trauma. Furthermore, the patient added that he has a habit of walking barefoot at home. During the period between the patient finding the lesion and visiting the doctor, the black line had been extending in an irregular linear fashion. It started at the base of the fifth toe and extended in a S-shaped direction towards the heel. Then, it turned 180° in an arc-shaped fashion and moved further towards toes, leaving a dusky grey-black, 8-shaped, serpiginous, linear eruption resembled CLM. It tracked within the skin for more than 10 cm (Figure 1a). On the initial cursory inspection, an evident, fine, superficial, black line was clearly observed through the skin surface at the leading end, which follow by a grey-black, 8-shaped linear eruption. When the black line was inspected under the dermatoscope, only a black-brown stripe could be clearly seen (Figure 1b), neither tunnels nor worms were observed. Summing up the above, a diagnosis of CPM was made. We performed a shallow incision at the advancing end, a slippery, straight, black line without follicle, was withdrawal in its entirety with a forceps. Under a light microscope, the black line was demonstrated to be a naked hair shaft, confirming the diagnosis of CPM. Complete remission without recurrence and sequelae was observed at three and a half months after extracting the hair (Figure 1c).

Table 1: Characteristics of the cases reported in the literature and our patient.

Patient	Year	Sex	Age (year)	Country	Site	Author	Length of hair (cm)
1	1957	F	19	USA	left ankle	Yaffee et al.	4
2	1960	F	1.25	USA	right sole	Schamberg et al.	2
3	1960	F	32	USA	Toe	Ronchese et al.	N

4	1975	M	2	Finland	right foot	Lehmuskallio et al.	0.9
5	1985	M	31	Japan	right groin to buttock	Takino et al.	3
6	1986	M	43	Japan	pubis to iliac crest	Ohta et al.	3
7	1990	M	39	Japan	near the navel	Takino et al.	3
8	1993	F	32	Japan	iliac crest	Ueda et al.	5.5
9	1994	F	31	Japan	left breast	Oka et al.	1.5 and 0.5
10	1994	F	21	Japan	pubis to left abdomen	Takagi et al.	N
11	2001	M	2	Japan	right outer sole	Shibuya et al.	N
12	2001	F	32	Japan	right groin to waist	Nakayama et al.	1.3
13	2002	F	31	Japan	left groin to lower back	Nakayama et al.	3
14	2005	M	55	Japan	left groin to lower back	Sakai et al.	4.5
15	2009	M	28	China	left sole	Luo et al.	3.5
16	2010	F	38	China	left foot	Luo et al.	N
17	2010	M	62	Japan	left foot	Arakawa et al.	2.5
18	2010	M	3	Korea	Feet	Kim et al.	2.2
19	2010	M	28	China	right cheek	Gao et al.	1.2
20	2012	M	0.5	China	left leg	Xie et al.	2.5
21	2012	F	2	Turkey	Foot	Yilmas et al.	NF
22	2013	M	46	Korea	Neck	Kim et al.	7
23	2015	F	30	India	Breast	Khare et al.	N
24	2015	M	42	India	Toe	Khare et al.	N
25	2016	M	2	Italy	right foot	Traniello Gradassi et al.	N
26	2016	M	0.67	India	scalp	Nagar et al.	N
27	2017	M	3	Italy	left foot	Traniello Gradassi et al.	N
28	2017	M	1.17	Mexico	Foot	Lopez et al.	NF

29	2018	M	39	Japan	Neck	Ishida et al.	0.2
30	2018	M	3	China	right foot	Li et al.	0.3
31	2019	F	39	Thailand	Neck	Ingkapiroj et al.	N
32	2019	M	25	China	left sole	present case	N
33	2020	F	32	China	left waist	Yang et al.	6
34	2021	M	1.17	China	sole of right foot	Yang et al.	4
35	2021	F	2.75	China	toenail	Yang et al.	3
36	2021	F	8.67	China	right sole	Yang et al.	2.5
37	2021	F	1.17	China	right toe	Yang et al.	0.5
38	2021	F	7.83	China	toe	Yang et al.	1
39	2021	F	7	Korea	axillae and neck	Nam et al.	0.03-0.3
40	2021	F	2	China	left sole	Jiang et al.	1.8

F, female; M, male; N, not described; NF, not found

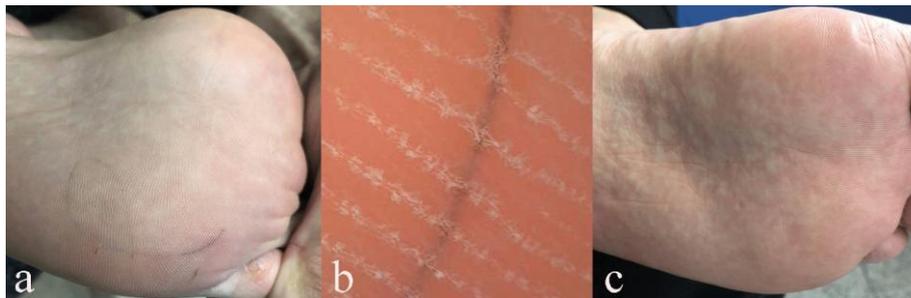


Figure 1: a) A dark line and, in continuity with it, a blurred dusky grey-black, 8-shaped, serpiginous, linear eruption on the left sole. b) Dermoscopy showed a black-brown stripe under the skin. c) The lesion diminished completely after removal of the hair.

Discussion

CPM is an uncommon disease caused by a hair shaft or fragment penetrate in the epidermis and occasionally in the middle dermis, which characterized by creeping eruption with a black line-like hair at the advancing end. Causative hair extraction led to a healing of the eruption [21]. From 1957 to the present, more than 40 cases were reported, but it should be certain that the actual number of cases must be more for some cases were unreported and the literature we have sought for was not all. Among the 40 cases we have found, 19 were female and 21 were male, accounting for 47% and 53% respectively (Figure 2a), which indicates no significant in incidence between male and female. The majority of cases have been reported from East Asian countries such as China, Japan and South Korea (Figure 2b). The locations involved included the ankle [3], sole [6,10,14], toe

[5,11], breast, inguinal fold [7] and neck [9,12]. The foot was the most commonly involved site. Reported lesion locations are mostly single sites, but it can be multiple in a few cases. Kim et al. [8] presented a 3-year-old boy with hairs on the soles of his feet, and Nam et al. [17] reported a rare case of multiple, embedded short hairs on the axillae and neck and named it ‘pili cuniculati multiplex’. The patients’ ages ranged from 6 months to 62 years, the mean age is 20.65 years, median age is 23 years. The characteristics of the cases, including ours, are summarized in [Table 1](#).

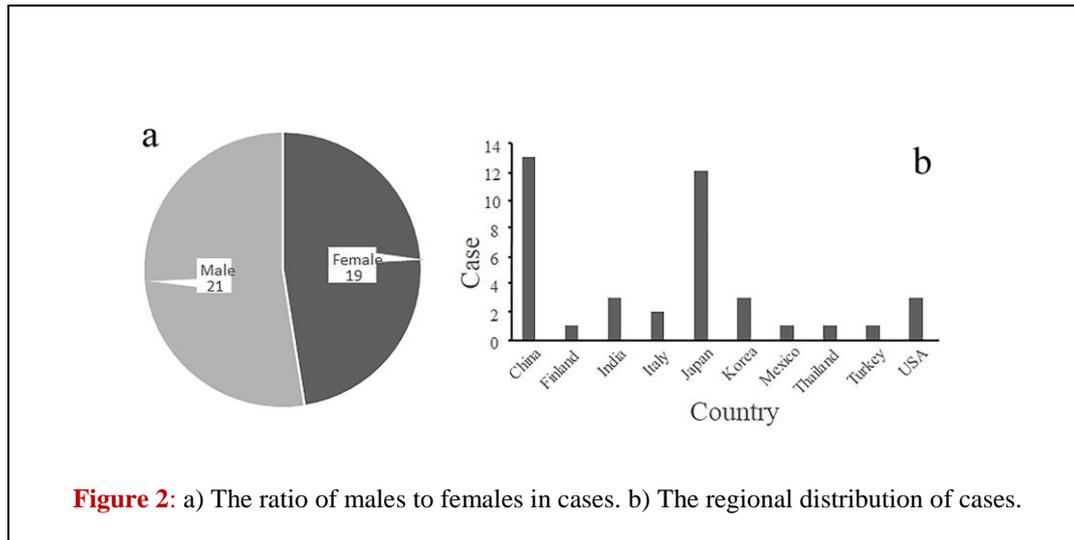


Figure 2: a) The ratio of males to females in cases. b) The regional distribution of cases.

The etiology of CPM still remains unclear. As no hair follicles have been observed in the published literature, the origins of the causative hair shafts or fragments are thought to be extrinsic. It is believed that the extrinsic sharp-pointed hair can penetrate the skin directly under the external force. The situation could happen easier when the skin is delicate (like the skin of an infant) or traumatic. When the tegmen of the laminated hair cuticle is just outside the skin, it can prompt the penetrating process like a barb and keep the hair from sliding out of the skin. Walking barefoot, soaked skin and frequent friction may be the predisposing factors [8,14,15]. In view of this, it is best to pull out the hair from the leading end, because it is easy to break it if against the direction of hair cuticle. Franbourg et al. [22] reported that Asian hair had the largest cross-sectional area with highest tensile strength among all ethnic groups, which may explain the fact that most cases were reported from East Asia. The embedding of hair in the skin and subsequent moving may be caused and maintained by biomechanical forces exerted by limb and body movement. The hypothesis has been supported by the fact that walking made the lesion extend more rapidly and the pain more evident in some reported cases [14,15]. The diagnosis of CPM depends on careful physical examination. The diagnosis can be made based on the typical manifestation of a serpiginous, slightly elevated, erythematous, linear migrating skin lesion, with obviously black line at the advancing end and accompanied by pain of varying severity. And it would be established if the extracted black line shows the hair structure under the microscope.

It is important to distinguish CPM from other conditions which manifest in creeping eruption via dermatoscopic and microscopic examination. Especially, a differential diagnosis with CLM is needed as the two conditions closely resemble each other. Firstly, compared with CPM, CLM can move in any direction and generally results in more serpiginous or tortuous tracts. Furthermore, lesions in CLM are extremely itchy and often accompanied by secondary dermatitis, whereas those in CPM may be either asymptomatic or slightly painful. Thirdly, a black

line can be seen in the head of CPM, while distinct tunnels and occasional worms in CLM. Finally, the histopathological features are inconsistent. Being identical to a foreign-body reaction, inflammatory cells consist of neutrophils, histiocytes and lymphocytes but no eosinophils, which are characteristic of CLM, can be observed in CPM [23]. Likewise, the conditions from intradermal growing hair and CPM should be differentiated from one another. In exceptionally rare instance, a hair grows inside the skin and burrows in the uppermost dermis rather than through the skin straightly, which described as different terms such as ‘cutaneous pili migrans’ [19], ‘creeping hair’ [24], ‘ingrowing hair’ [25] and ‘intradermal growing hair’ [26]. Among these, the term of ‘intradermal growing hair’ is thought to be a better description of the condition [26]. The two conditions were once confused with each other for the extremely similar manifestation of movable, black, thin linear eruption. However, these eruptions vary in their origin and characteristics. The source of CPM is extrinsic, usually found crawling in the epidermis, and the shaft is clearly visible through the skin, while hair that grows in the skin is internal, usually found burrowing in the dermis, and is not readily visible. Another important point is that the length of black line in CPM remains constant, but it can get longer due to the hair growing in the counterpart.

Conclusion

CPM is a rare and interesting condition which easy to diagnose and cure. Differential diagnosis includes CLM and intradermal growing hair. Close examination is critical to diagnose, and pulling the hair out from the leading end in a non-invasive or minimally invasive way with a forceps can heal completely. Physical examination should be emphasized while patients presenting with creeping skin eruption.

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