Letter to the Editor

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Trichoteiromania: An Atypical Case Associated with the Claude Bernard Horner Syndrome

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Dear Editor,

Trichoteiromania is a self-inflicted hair disorder that consists in the act of rubbing the scalp with fracturing of the hair shafts [1]. Freyschmidt-Paul et al. [2] proposed the term trichoteiromania, because "teiro" is a Greek derivation for "I rub."

We describe a 56-year-old woman who compulsively rubbed her scalp hairs, resulting in a noncicatricial alopecia patch and fractured hair shafts, evidenced by the presence of trichoptilosis in trichoscopy and optical microscopy.

Clinical Case

A 56-year-old woman presented with a 3-year history of broken scalp hairs restricted to the left frontoparietal region. About 6 months prior to this, she presented with left-sided myosis, upper eyelid ptosis, and anhidrosis, and was diagnosed with the Claude Bernard Horner syndrome secondary to C6–C7, C7–T1 and T6–T9 syringomyelia, proven by nuclear magnetic resonance. She initially denied itching localized to the affected scalp. However, after reporting that itching caused the hair disorder, she realized that she actually rubbed the spot. The patient denied cutting or pulling scalp hairs. There was no past history of eczema, seborrheic dermatitis, psoriasis, or alopecia or any other significant past medical history.

Physical Examination

The left frontoparietal region showed broken hairs measuring 1–2 cm. There was preservation of normal-length hairs at the margins (Fig. 1). The pull test as well as the tug test were negative.

Trichoscopy (Dermlite DL3N)

The following results were seen with trichoscopy:

- *Scalp:* absence of scaling, erythema, atrophy, and white, black or yellow dots (Fig. 2).
- *Shafts:* trichoptilosis – longitudinal splitting of the distal end (Fig. 3).

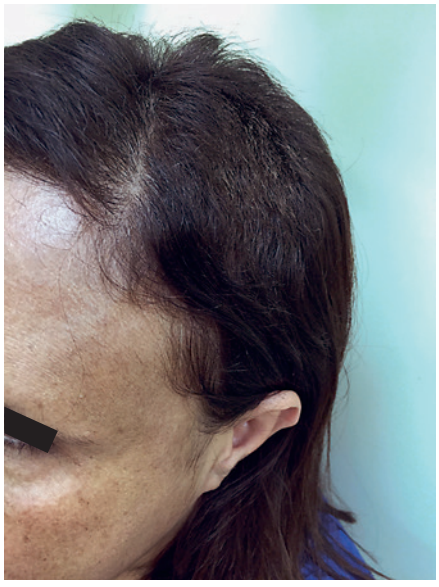

Fig. 1. Left frontoparietal region with broken hairs measuring 1–2 cm. Preserved hair shafts at the margins.

Fig. 2. Trichoscopy of the scalp: absence of scaling, erythema, atrophy, and white, black or yellow dots.

Fig. 3. Trichoscopy of the shafts: brush-like end.

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Optical Microscopy

Optical microscopy showed trichoptilosis, i.e., longitudinal splitting of the distal end of the hair shaft (Fig. 4).

Discussion

Trichoteiromania is a self-inflicted hair disorder that consists in the act of rubbing the scalp with fracturing of the hair shafts. It is part of compulsive hair disorders, of which trichotillomania is the best known. However, there are others less known, such as trichotemnomania, trichoteiromania, trichophagia, and trichocryptomania [1]. Trichoteiromania is characterized by the presence of short shaft hairs, 1–2 cm long, and trichoscopy shows longitudinal splitting of the hair shaft, called trichoptilosis. The disorder can eventually be precipitated or aggravated by a scalp pathology, such as eczema, seborrheic dermatitis, or dermatophytosis [1].

Differential diagnoses in our patient were regrowing alopecia areata, trichotillomania, trichotemnomania, tinea capitis, and trichorrhexis nodosa.

The most characteristic trichoscopic features of alopecia areata are yellow dots, black dots, exclamation mark hairs, tapered hairs, broken hairs, vellus hairs [3]. Regrowing alopecia areata shows normal end hair shafts.

Trichotillomania trichoscopic features include broken hairs at different lengths, short hairs with trichoptilosis, coiled hairs, exclamation mark hairs, and black dots [4]. Recently, new characteristic trichoscopic features of trichotillomania have been described, including flame hairs, the V-sign, hook hairs, hair powder, and tulip hairs [5].

Trichotemnomania is hair loss due to cutting or shaving in the context of a compulsive disorder. The hair is usually cut with scissors or by shaving and the diagnostic key is the presence of follicle openings with filled hair shafts within a healthy-looking scalp [6].

Tinea capitis trichoscopic aspects of the hair shaft are comma hair, corkscrew hair, bar code-like hair, zigzag hair, broken hair, and black dots [7].

Trichorrhexis nodosa is an easily breaking hair with nodular structures along the hair shaft. Trichoscopy shows nodular thickening of the hair shaft, which appears light in the dark hair. Eventually, the break of hair shaft leaves a rounded or brush-like end [8].

Due to the proximity of the cutaneous and neural events of the patient reported, it was decided to investigate a possible relation between them.

Syringomyelia refers to a cavitation of the spinal cord and the clinical picture varies according to the site affected. It commonly reaches the cervical spine, which can cause sensory loss on the shoulders and radicular pain of the upper limbs and neck. When it reaches the Th1 vertebra, it can cause the Claude Bernard Horner syndrome, as happened in this case.

In the Claude Bernard Horner syndrome, the sympathetic pathway is reached, with myosis, palpebral ptosis, and ipsilateral anhidrosis [9].

In order for syringomyelia to compromise the frontotemporal area of the scalp, innervated by the trigeminal nerve, the lesion should be higher, where the bulb is.

In contrast, the Claude Bernard Horner syndrome reaches the sympathetic nerves, causing anhidrosis, the trigger most likely being associated with the pruritus of the patient. To clarify this situation, the starch-iodine test was performed on the forehead and scalp (frontoparietal), confirming hypo/anhidrosis on the left side (Fig. 5, 6).

A Mayo Clinic study selected 227 patients with localized cutaneous pruritus, burning and paresthesia, with no apparent cause to assess the sweating pattern, in order to attribute these sensations to short nerve fibers, which also control sweating. At the end of the study, 149 patients (66%) had abnormal results and 119 (80%) had anhidrosis at the sites where they had symptoms, suggesting that idiopathic pruritus should be tested to search abnormal sweating [10].

Color version available online

Fig. 4. Optical microscopy: brush-like end.

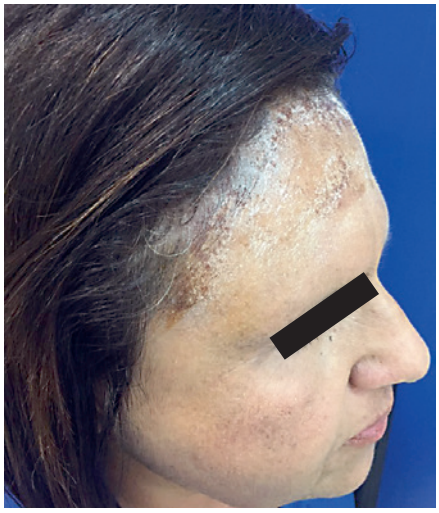

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Fig. 5. Iodine-starch test on the right side: noted dark blue-black discoloration at the side of normal sweating.

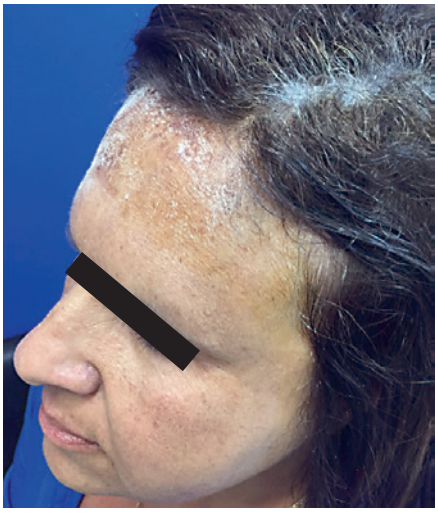

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Fig. 6. Iodine-starch test on the left side: without color change, due to hypo/anhidrosis.

Shimoda et al. [11] evaluated patients with lichen amyloidosis and showed that they had hypo/anhidrosis, suggesting that skin dryness would be the starting point of pruritus.

In the presence of pruritus of the scalp with absence of cutaneous abnormalities and psychiatric disorders, peripheral nerves and cervicothoracic spine abnormalities should be investigated [12].

Statement of Ethics

The authors have no ethical conflicts to disclose. The patient's consent has been obtained.

Disclosure Statement

The authors have no conflicts of interest to disclose.

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