Trichoteiromania: Good Response to Treatment with N-Acetylcysteine

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Established Facts

- Lichen simplex chronicus arises from excessive primary scratching; the scalp is one of the most commonly affected sites.
- The treatment of lichen simplex chronicus on the scalp, also known as trichoteiromania, is difficult and it is often refractory. Potent topical glucocorticoids are frequently used, but their success is variable.
- N-acetylcysteine (NAC) has been used in the treatment of some obsessive-compulsive dermatological conditions, and this is effective in adult patients with trichotillomania and skin picking disorder.

Novel Insights

- NAC might be useful in the control of the urge to scratch and rub the scalp, and lead to the regrowth of hair of the lichenified plaques, which result from trichoteiromania.
- To our knowledge, we report the first case of trichoteiromania successfully treated with oral NAC monotherapy.

Keywords

Trichoteiromania · Lichen simplex chronicus · Scalp · N-acetylcysteine

Abstract

Lichen simplex chronicus on the scalp, also known as trichoteiromania, can be difficult to manage, as the therapeutic options are limited to topical or intralesional glucocorticoids. We describe a patient with trichoteiromania, presenting three lichenified pruriginous plaques on different regions of the scalp, associated with fracture and loss of hair shafts. Prior treatment with potent topical glucocorticoids was ineffective. However, treatment with oral N-acetylcysteine (NAC) 1,200 mg/day resulted in complete hair regrowth within 16

weeks. NAC is a safe drug with a good tolerance profile that could be a therapeutic option for patients with trichoteiromania. The potential of NAC has not been completely elucidated, thus more studies will be necessary to confirm its efficacy in the long term for some psychodermatological conditions.

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Introduction

Trichoteiromania is the term used to refer to the compulsive action of scratching and rubbing of the scalp, which results in fracture of the hair shafts [1, 2]. The standard treatment includes potent topical glucocorticoids or intralesional glucocorticoids. However, due to the “itch-

Fig. 1. Well-defined hyperchromic plaques with regular borders and broken short hairs (a–c). Trichoscopy shows erythema, peri- and interfollicular scaling and broom hairs (d–f). In addition, hyperpigmentation and lichenification in the right parietal lesion (d) as well as hematic crust in the left parietal-occipital lesion (e).

rub/scratch-itch” cycle, the success is variable [3, 4]. Although cognitive behavioral therapy oriented to insight may be useful [5], many patients refuse this, or find it difficult to maintain in the long term. Thus, other therapeutic strategies are necessary. N-acetylcysteine (NAC) has been used with good results in the treatment of obsessive-compulsive disorders (OCD), and more specifically in some psychodermatological conditions, such as onychotillomania, skin picking disorder, prurigo, and trichotillomania [6–14].

Case Report

A 47-year-old man presented with an 8-year history of pruriginous plaques on the right parietal region. After 4 years, two new pruriginous plaques arose, in the occipital and left parietal-occipital regions, associated with hair loss in these regions (Fig. 1a–c). There was no medical history of primary diseases of the scalp such as psoriasis, seborrheic dermatitis, eczema or any other. The patient admitted rubbing his scalp for at least 3 h a day in the regions mentioned and denied pulling or cutting his hair. On physical examination, three well-defined hyperchromic plaques were observed with regular borders and broken short hairs. Under trichoscopy (DermLite DL4), all lesions presented erythema, peri- and interfollicular scaling and broom hairs. (Fig. 1d–f).

During the last year, treatment had been carried out with clobetasol cream twice a day, without improvement. The patient rejected psychotherapy and did not wish to use topical treatments again. Treatment was begun with oral NAC 1,200 mg/day. There was partial improvement at 8 weeks and complete hair regrowth at 16 weeks in all patches. (Fig. 2a–f). According to the patient,

there was a significant decrease in the impulse to scratch. The response to the treatment was maintained at the 30 weeks of follow-up. No adverse effect associated with the medication was reported.

Discussion/Conclusion

Lichen simplex chronicus is a skin disorder characterized by lichenification as a result of excessive primary scratching. The scalp is one of the most affected sites and itching is the main symptom. The peak of incidence is between 35 and 50 years, and it is more frequent in women. The persistence and progression of the lesions is correlated with the scratching and rubbing activity [3].

In 2001, Freyschmidt-Paul et al. [1] suggested the term trichoteiromania to describe the self-inflicted cases of compulsive rubbing of the scalp with fracturing of the hair shafts. Clinically, it presents as single or multiple lesions, which are irregular, oval or polycyclical, well-defined, with scaling and hair loss or fracture of hairs [15]. Broom hairs have been described as a trichoscopic feature of trichoteiromania. They represent broken hair shafts longitudinally split into 2 or 3 parts. The distal tips may have additional splitting [16]. They have also been described in trichotillomania [17]. Nevertheless, the clinical history and the presence of lichenification in one of the lesions favor the diagnosis of trichoteiromania in this case. In our experience, a patch of alopecia presenting

Fig. 2. Treatment with oral N-acetylcysteine monotherapy at a dosage of 1,200 mg/day in a 47-year-old man diagnosed with trichoteiomania, demonstrating complete hair regrowth at 16 weeks of therapy in all patches. Clinical examination (**a–c**) and trichoscopy (**d–f**).

clusters of broom hairs under trichoscopy is highly suggestive of trichoteiomania.

Management of patients with lichen simplex chronicus is difficult and the disorder is often refractory. Most commonly reported treatments are topical and intralesional glucocorticoids, but with variable success. Anecdotal reports also mention the use of topical capsaicin, tacrolimus, and doxepin hydrochloride. Psychotherapy is recommended as an adjuvant treatment [3, 18, 19].

NAC is a thiol compound, which has been used in psychodermatological conditions such as onychotillomania, skin picking disorder, prurigo, and trichotillomania [6]. Grant et al. [7] evaluated the efficacy of NAC from 1,200 to 2,400 mg/day in a double-blind, placebo-controlled study on 50 adult patients with trichotillomania, and they observed a significant reduction of hair pulling compared to placebo. Similarly, there are several case reports of patients with trichotillomania successfully treated with oral NAC [8–12]. The mechanism of action of NAC in the treatment of OCD is possibly due to its action on the glutamate system: glutamatergic hyperactivity leading to excitotoxicity and oxidative stress has been implicated in the pathogenesis of OCD [13]. NAC is converted to cysteine in the liver, which in turn is converted to glutathione, which has antioxidant effects [6, 13]. Residual cysteine in the systemic circulation is able to pass through the hematoencephalic barrier and reach the brain, where it will be oxidized to cystine. Subsequently the extracellular cystine is exchanged for intracellular glutamate through

the cystine-glutamate antiporter, principally located in the glial cells. The glutamate now in the extracellular space activates the mGluR2/3 receptors. These receptors are predominantly presynaptic and negatively modulate glutamatergic neurotransmission [13]. Additionally, it has been demonstrated that NAC alters the level of dopamine in neurons [6]. The decrease in the levels of glutamate, being the main excitatory neurotransmitter, in the nucleus accumbens would explain the usefulness of NAC in the control of compulsive behavior [6, 13].

In conclusion, we report a case of trichoteiomania presenting with hair regrowth after treatment with NAC. The obsessive-compulsive nature of this disorder may explain the success of therapy with NAC in this case. Additional reports with a larger number of patients are needed in order to confirm our findings. Finally, we suggest readers to consider trichoteiomania as one of the differential diagnosis in patients presenting with patchy alopecia.

Statement of Ethics

A written consent was obtained from the patient.

Disclosure Statement

The authors have no conflicts of interest to declare. There were no funding sources for this work.

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