



Trichotilliosis Totalis and Trichotilliosis of the Wig as a Consequence of Long-lasting Trichotillomania

Mehdi Iskandarli, MD¹
Banu Yaman, MD²
Fezal Özdemir, MD²

¹ HB Güven Klinik, Department of Dermatology and Venereology, Baku Azerbaijan

² Ege University Faculty of Medicine, Department of Pathology, Izmir, Turkey

Correspondence:

Mehdi Iskandarli, MD
Dermatology and Venereology
Department, HB Güven Klinik, Baku,
Azerbaijan, AZ1108

E-mail: info@mehdiiskandarli.com
Tel: +994 55 305 25 20

Background: Trichotillomania usually evolves in patchy form. Clinicians may confuse patchy form of alopecia areata with trichotillomania because of their similar clinical manifestation which consequently may lead to a misdiagnosis. Alopecia totalis rarely can be confused with trichotillomania. Here we report a case, where trichotillomania shows similar clinical appearance with alopecia totalis.

Objective: To demonstrate the rare variant of trichotillomania which share similar clinical features with alopecia totalis. In order to escape from misdiagnosing and mistreating of trichotillomania, we recommend a new nomenclature and classification for hair pulling disease.

Material & methods: Case discussion and brief review.

Results: Total hair loss and patchy alopecia of the wig due to trichotillomania, very rarely reported in the literature. Present case and literature review demonstrate that psychogenic total hair loss of the scalp and alopecia of the wig, maybe sign of untreated and long-lasting trichotillomania.

Conclusion: Terms like "trichotilliosis areata" and "trichotilliosis totalis" could be used in order to avoid confuseness of alopecia areata and alopecia totalis, respectively.

Keywords: Trichotillomania, Trichotilliosis Totalis, Trichotilliosis Areata, Trichotilliosis of the Wig, Alopecia Totalis, Alopecia Areata.

Introduction

Hair pulling and skin picking are body-focused repetitive behaviors induced by preceding anxiety and increasing sense of tension.[1] Above mentioned behaviors is a part of psychocutaneous disorders like trichotillomania (TTM) and dermatotillomania respectively, which were listed in the section of obsessive-compulsive and related disorders (OCRD) in DSM-5. [1] In patients with TTM, preceding anxiety and increasing sense of tension leads to consciously or unconsciously hair pulling from own scalp, eyebrow, eyelash, axillar and pubic region which may endure for months and years.[1,2] Some patients may pull hairs from pets, dolls, sweaters and carpets. [1] Scalp most common affected region in

TTM. Due to continuous hair pulling from one side or multiple sides of scalp, patchy alopecias may evolve in these patients.[1,2] Most common pulling sites on scalp are vertex and parietal region.[3] Usually, hairs are in different length and configuration in TTM patients.[2,3] Because of non-inflammatory nature of the disease, redness, desquamation and any other symptoms are absent in these patients. Therefore, TTM can be confused with alopecia areata (AA), in which macroinflammation and any other symptoms also is not presents.[2,3] Alopecia totalis (AT) rarely can be confused with TTM. Because, total hair loss due to hair pulling, is not common in TTM. Here we report an extreme case of TTM, in which AT considered in differential diagnosis.

Case Presentation

Nineteen-year-old female patient referred to dermatology department with massive hair loss on scalp. Patient's hairs started to loss when she was twelve-year-old. Hair loss was started when she changed her school to another one. Patient was on wig, at first meeting with a dermatologist. On physical examination of scalp, massive hair loss and hair shafts in different length was found. Hairs of eyebrow and eyelash was almost completely pulled out. Furthermore, single large alopecia patch was found at vertex of the has got a total hair loss. (fig. 1). Genital and axillar hairs was intact. Nail examination revealed severe onychophagia. On trichoscopic evalution, coiled hair shaft, flamme sign, v-sign and black dots were detected on scalp (fig. 2). Histopathologic evaluation of skin sample taken from scalp, revealed noninflammatory empty anagen phase hair follicles (fig. 3). Patients routine blood test was normal. Ferritin and Zn level was normal. Thyroid gland abnormalities was absent. All autoimmune markers was negative. Patient at the end, referred to psychiatrist and sertraline with risperidon initiated for the treatment of underlying psychiatric disorder.

Discussion

Underlying psychiatric disorders like OCRD may cause both, AA and TTM. In presented case, patient referred to dermatology unit with preliminary diagnosis of AT. However, following detailed examination of the wig and scalp, TTM was considered at first. Patient with AT apperance, diagnosed as a severe TTM following trichoscopic and histopathologic examination. Exclamation mark on trichoscopy, swarm of bee on histopathology and autoimmune markers on blood examinations was negative. So, AT ruled out by these results. Furthermore, on trichoscopy flamme hairs, coiled hairs, V-sign and black dots was positive for TTM. This patient suffered from TTM for seven year. First time she referred to a specalist when she already has got a total hair loss. Patient misdiagnosed and treated as a AT several time, before hospitalisation in our department. Probably, the severity of TTM is time dependent in present case. Long-lasting anxieties, led to long-lasting trichotillar repetative behaviors, as a consequence total hair loss was evolved. Once total hair loss established following continous anxieties and hair pulling tic, this patient started to drag out hairs of the wig from the vertex untill tonsure pattern alopecia evolved. Later, patient confessed that first hair pulling site was vertex. The first hair pulling site of the wig was also vertex. Therefore, patients with AT, wig examination should be implemented also, for diagnostic purpose. Because, in severe and long-lasting TTM cases, trichotillosis of the wig may evolve following the total trichotillar hair loss, which clinically could be similar to AT. In the literature, reports regarding total hair loss due to TTM is very limited. However, TTM of the wig, is an exceptionally rare case and never reported in the literature. In one report, patient suffered from TTM for four year, another one for nine year.[4] In both patient alopecia initially was patchy type, then hair loss gradullay became totally. So, patient should be diagnosed and treated in early patchy stages of TTM, otherwise long-lasting TTM can cause total trichotillar hair loss,



Figure 1.
Trichotillosis totalis and trichotillosis of the wig in tonsure pattern.

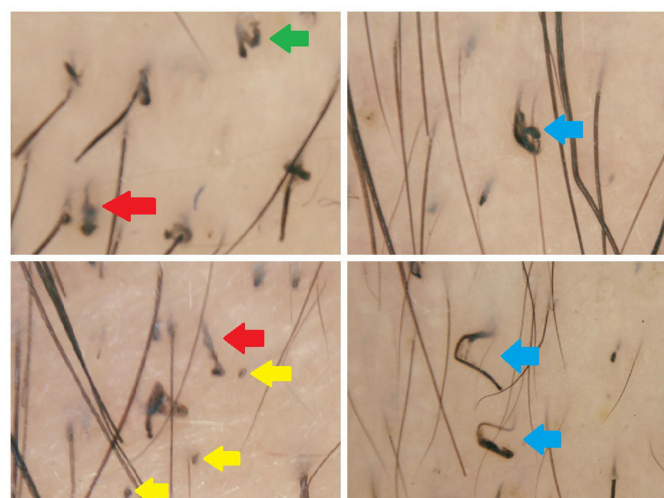


Figure 2.
Flamme sign (red arrow), V-sign (green arrow), coiled hairs (bule arrow) and black dottes (yellow arrow).

even trichotillosis of the wig. In present case, patient diagnosed TTM and treatment against underlying psychiatric disorder was initiated, seven years after the first signs of hair losses.

First time Dimino-Emme et al. described the "Friar Tuck sign" or tonsure pattern of hair loss in TTM patients which is a single large alopecia patch at the vertex.[5] In present case, alopeci patches also was started in a tonsure pattern which later generelised due to long-lasting hair pulling, according to confession of the patient. "Friar Tuck sign" on the wig confirm the righteous confession of patient. Unfortunately, we lost the

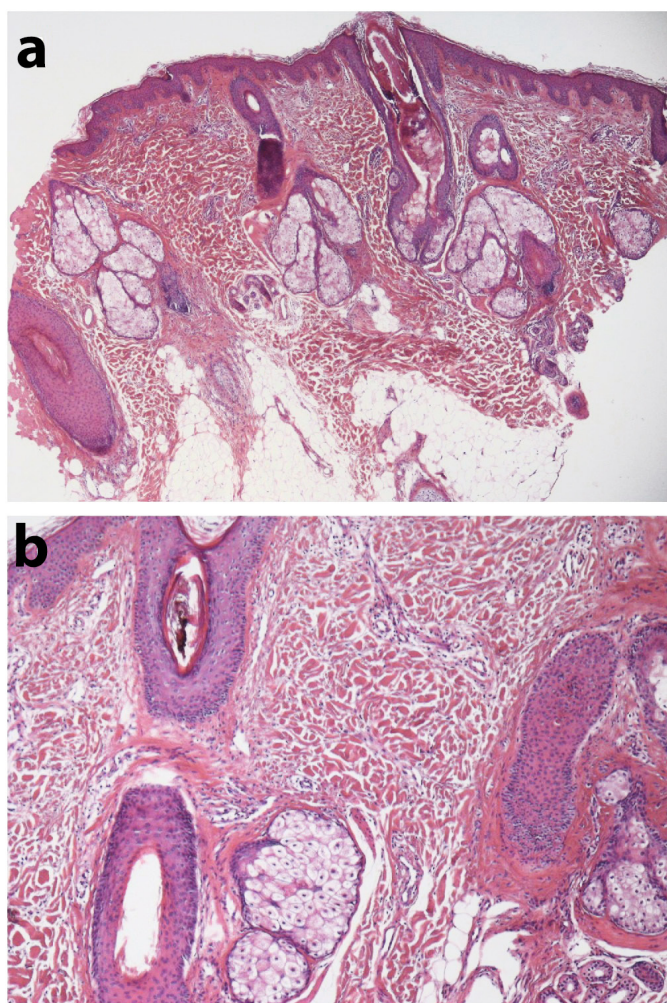


Figure 3.

Three anagen and one catagen phase hair follicles on noninflammatory base H&E $\times 40$ (a). Empty anagen phase follicle H&E $\times 100$ (b).



Figure 4.

Zoomed macropicture of tonsure pattern alopecia of the wig. Hair shafts in different length is detectable.

tillotic” as an adjective could be used in the terminology like “Trichotillotic alopecia” as in “Herpetic gingivostomatitis” or “Neurotic person” but it doesn’t open the whole spectrum and severity of the diseases. Therefore, we think that trichotilliosis is more relevant term. The term TTM should be considered as psychiatric conception like ‘kleptomania’ and ‘pyromania’. However, trichotilliosis (trix+tillein without mania) should be accepted as a dermatological concept of TTM syndrome. In other word, underlying anxieties and increasing sense of tension, beside trichotillotic body-focused repetitive behaviors together making the OCRD, so called TTM. Probably, the severity of trichotilliosis depends on severity and duration of obsession. At the early stages of TTM, patchy form of alopecia or trichotilliosis areata evolves. Trichotilliosis areata is a most common form hair loss in TTM. Trichotilliosis totalis is massive hair loss which is uncommon subtype and probably it develops in long-lasting untreated TTM patients like in presented case (Table 1).

Conclusion

In conclusion, by this article we are trying to demonstrate the extremely rare case of TTM with a wig involvement. In present case, patients diagnosed as TTM after seven year later. Probably, due to untreated and long-lasting course of the disease caused development of trichotilliosis areata of tonsure pattern which consequently generalised and became trichotilliosis totalis and trichotilliosis of the wig with a positive friar tuck sign. Patients with trichotilliosis totalis could be confused with AT and mis-treated. Therefore, patient with AT on wig, have to be carefully evaluated and wig examination also should be done in order to exclude severe trichotilliosis totalis, since OCRD can cause both, AT and TT by different pathogenic pathways.

chance to evaluate the wig trichoscopically in order to find the clues for confirmation of TTM. However, hair shafts in different length is visible when zooming the macropicture of the wig (fig. 4)

Following the brief review of the literature regarding total hair losses in TTM, we recommend to separate the terms TTM and trichotilliosis in order to better understand the severity dependent clinic of the disease. TTM derived from greek: trix (hair), tillein (to pull) and mania (madness). First time this term coined in 1889, by french dermatologist François Henri Hallopeau.[6,7] Aljabre in his article recommended different names for TTM, like trichotilliosis, trichotillotic and tic trichotilliosis.[8] Horenstein et al. used the term trichotilliosis instead of TTM, in their article.[9] Castelo-Soccio used TTM and trichotilliosis as a synonym terms.[10] Shelleh et al. preferred to accept the term “trichotillotic” in their article.[11] Probably, the meaning of that term is a “hair pulling tic”. However, the term “trichotillotic” at first glance, pronouncing like an adjective of unknown noun, in which adjective alone don’t make a sense. The word “tricho-

Table 1. Recommended clinical subtypes of hair loss in TTM.

Clinical subtypes	Explanation
Trichotillosis areata	Patchy alopecia. Patches start usually from vertex and parietal region. Extensive patchy alopecia of vertex correspond to tonsure pattern of TTM. Early stages of TTM maybe multipatchy and very oftenly can be confused with AA
Trichotillosis totalis	Total hair loss of the scalp. Rare form TTM. It develops in long-lasting and untreated TTM patients. Can be confused with AT.
Trichotillosis of the wig	Patchy alopecia of the wig. The location of the patches probably correspond to initial hair pulling site on scalp. Could be sign of severe TTM
Trichotillosis universalis	Never reported form. Theoretically, in a long-lasting anxieties patient may pull out all terminal hairs, including scalp, eyebrow, eyelash, axilla and pubis

TTM - trichotillomania, AA - alopecia areata, AT - alopecia totalis.

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