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Tinea Capitis in Two Sisters of a Wooly Hair Family

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Abstract

Observation: Wooly hair is usually present at birth or infancy with a genetic linkage of autosomal dominant or recessive. Hair is curly, thick and often heavily pigmented. This condition has been reported with eye, teeth, cardiac anomalies. Also, keratosis pilaris atrophicans, ichtiyosis and deafness, palmoplantar keratoderma and Noonan syndrome may accompany wooly hair. We report two sisters with wooly hair, simultaneously developed an inflammatory tinea capitis (kerion). Our patients have neither a systemic disease nor eye, dental and other skin disorders. In their family; mother, two sisters, and one brother of them have also wooly hair without any other clinical associations. To our knowledge, this is the second, describes the association of wooly hair with tinea capitis. However, in the first report, mother and her son, also had keratosis follicularis spinulosa decalvans. As a result, presence of tinea capitis in both patients may be explained by the enhanced susceptibility to fungal infection in keratinizing disorders.

Introduction

Wooly hair presents fine, tightly curled hair. Microscopically, the hairs are tightly coiled. This abnormal variant of the hereditary condition appears usually as a solitary problem. Also, classified into 3 variants: wooly hair nevus, autosomal dominant hereditery, and autosomal recessive familial variant [**1, 2, 3**].

Dermatophytes have ability to form molecular attachments to keratin and use it as a source of nutrients allows them to colonize keratinized tissues, including the stratum corneum of the epidermis, hair, nails. Resistance factors to the colonization of fungi composed of UV light, variation in temperature and moisture, and fungistatic fatty acids and sphingosines produced by keratinocytes. Spores have to germinate and penetrate the stratum cor-

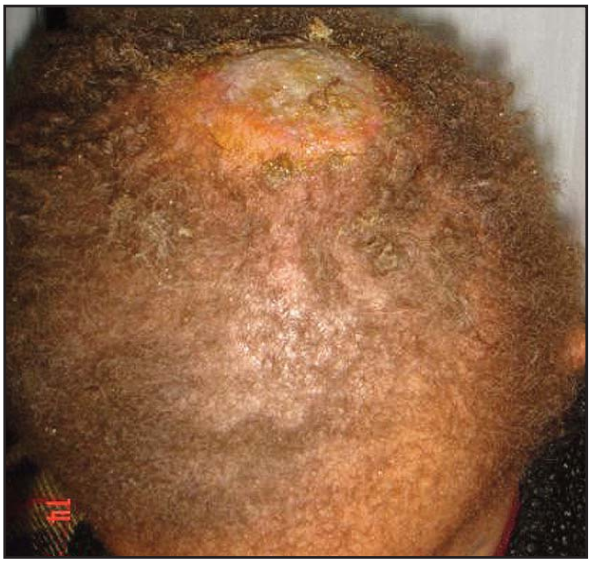

Figure 1. A boggy mass studded with broken hairs and follicular orificies oozing with pus on left parietal region of patient (Case 1)

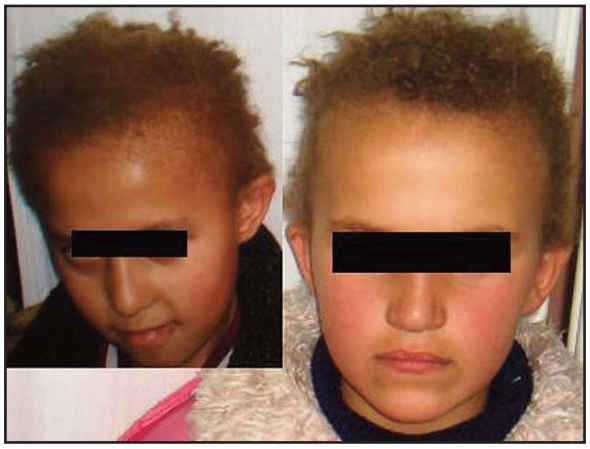

Figure 2. Two sisters

neum at a rate faster than desquamation. Trauma and maceration also faciliate penetration and degree of inflammation is highly dependent on immune system. Genetic immunological predispositions and also genetic differences of keratins affect the ability of a fungus to attach to the stratum corneum [**4, 5, 6, 7**].

We report here two sisters with wooly hair who developed simultaneously an inflammatory tinea capitis.

Case Reports

Case 1

A 10-year-old girl was referred to our clinic for evaluation of a wound on her head. The lesion beginned three weeks ago, as a small erythematous swelling and than spreaded. Dermatological examination showed a boggy mass studded with broken hairs and follicular orificies oozing with pus on left parietal region of her head (**Figure 1**). The remainder of the scalp showed curly, thick, heavily pigmented, and unmanageable hair; this aspect had been present since birth. Also bilateral cervical lympadenopathy were examined. Eyebrows and eyelashes were present and appeared normal. No additional clinical manifestation was observed in thorough dermatological examination including entire integument and oral mucosa. Cardiological and ophthalmological examinations revealed normal findings. No presence of any syndrome was observed during physical examination. Mycologic examination of the hairs from the lesion revealed ectothrix dermatophyte infection. Fungal culture of specimens did not identify any species of fungi.

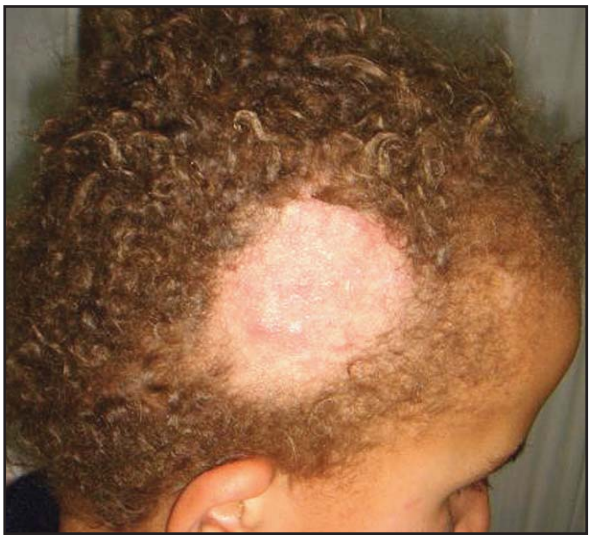

Figure 3. An erythematous and indurated plaque with alopecia on the right temporal region of her head (Case 2)

The examination of hair shaft carried out by light microscopy demonstrated tightly coiled hair.

Family history of the patient was remarkable. Her mother and brother, three sisters and also, her maternal uncle and aunt had wooly hair. One of her sister was referred together with her and presented similiar clinical findings (**Figure 2**).

Treatment with topical dressings and terbinafine at a dosage of 125 mg daily for one month [**6**] produced cure in our patient.

Case 2

A 13-year-old girl was referred to our dermatology clinic for evaluation of a wound on her scalp. Dermatological examination revealed an erythematous and indurated plaque with alopecia on the right temporal region of her head (**Figure 3**). The medical history, physical and mycological examinations were similiar with her sister, as described in Case 1. Dermatological examination of entire integument including all nail and cuticle was normal. Normal ocular findings were found in ophthalmological examination. Cardiological examination revealed normal findings. Also no clinical manifestation was observed to suggest any syndrome associates with wooly hair.

At follow-up, the patient was successfully treated with a 4-week course of terbinafine 250 mg/day [**6**].

Discussion

Tinea capitis is most commonly found in children ages 3 to 14 years; it is uncommon in adults [**5**]. Transmission is increased with dec-

reased personal hygiene and low socioeconomic status. Asymptomatic carriers are common, making tinea capitis difficult to eradicate [**7**]. It is known that telogen hair do not get infected by fungus. And, only newly formed keratin is suitable for fungal growth and as this is available only at the outer margin of the keratogenous zone, At this zone, fungal growth equals the rate of new keratin being formed at Adamson's fringe. As long as the fungus maintains the same rate of growth as that of the hair shaft the infection persists [**4**].

Wooly hair, appears as part of a syndrome such as *Carvajal* syndrome or *Naxos* disease in some cases [**2**]. Both diseases are characterized by cardiomyopathy, palmoplantar keratoderma and wooly hair, and they are caused by mutations in the desmoplakin and plakoglobin genes, respectively. As yet, however, the pathogenesis of hereditary wooly hair remains largely unknown [**1, 8**].

Two sisters described in this report suggest an association between inflamatorry tinea capitis (kerion) and wooly hair. Abnormal keratinization in wooly hair may play a role in the in patogenesis of tinea capitis. The two siblings described herein live together and also have education in the same boarding school. It is not clear whether the patients infected in house or in school. Our patients did not give definite knowlodges about the probable "tinea capitis" cases in their school. No member in the same household was presented with tinea capitis simultaneously. But they were living in a village with low socioeconomic status. Family have low income from animal breeding. A probable transmission of a zoophilic species may explore the cause of infection. Negative results of fungal cultures may explained by identifying difficulties of zoophilic fungi.

To our knowledge, this is the second, describes the association of wooly hair with Tinea capitis. However, in the first report [**3**], patients also had keratosis follicularis spinulosa decalvans (KFSD). Simultaneously an inflammatory tinea capitis developed in a son and his mother in whom KFSD occurred in association with wooly hair. In addition, various dental anomalies, were present in the child. The concomitant onset of inflammatory tinea capitis in both patients have been explained by the enhanced susceptibility to fungal infection in keratinizing disorders. In an another study [**9**], three teenage siblings with confluent and reticulated papillomatosis were described. Two of the three patients had confirmed tinea versicolor, with positive potassium hydroxide scrapings, in association with this entity. In this largest series of siblings with confluent and reticulated papillomatosis, family members had both confluent and reticulated papillomatosis and tinea versicolor. Investigators suggested that keratinizing disorders. in confluent and reticulated papillomatosis may be linked to tinea versicolor.

As a result, we think, presence of tinea capitis in both patients may be explained by the enhanced susceptibility to fungal infection in keratinizing disorders. Further studies exploring immunological and molecular mechanisms of keratin and the invasion of dermatophytes are needed.

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