

Pathologic grooming (acne excoriee, trichotillomania, and nail biting) in 4 generations of a single family

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INTRODUCTION

Trichotillomania (hair pulling disorder), excoriation (skin-picking) disorder, and other body-focused repetitive behavior disorders (BFRBDs) have received increasing attention in the psychiatric nomenclature. Trichotillomania was introduced in the Diagnostic and Statistical Manual of Mental Disorders (DSM) in DSM-III-R in 1987, whereas skin-picking disorder was introduced in the most recent edition of the manual, DSM-5.¹ DSM-5 includes both disorders and also refers to other less well-studied BFRBDs, such as nail biting, in a new chapter on obsessive-compulsive and related disorders.² A somewhat similar approach is likely to be taken in the next revision of the International Classification of Diseases.³

These disorders, which also represent pathologic grooming,⁴ may be particularly prevalent in dermatologic practice where they are sometimes precipitated by skin conditions such as acne (acne excoriee).⁵ Fortunately, there have been important advances in understanding the phenomenology and psychobiology of these conditions in recent decades. In particular, studies find that these are prevalent conditions that are associated with significant impairment and reduced quality of life and are responsive to specific pharmacotherapeutic and psychotherapeutic interventions.^{6,7} Studies emphasize that there is significant comorbidity between these conditions and that they may well share underlying psychobiological mechanisms, including familial and genetic risk factors.^{6,8}

Further work is needed to understand the familial and genetic risk factors for the BFRBDs. Very few gene association studies have been undertaken, and

Abbreviations used:

BFRBDs:	body-focused repetitive behavior disorders
DSM:	Diagnostic and Statistical Manual of Mental Disorders

it is unclear whether genetic variants that are associated with BFRBDs are specific for these conditions or are associated with vulnerability to a larger group of obsessive-compulsive and related disorders.^{9,10} We present a South African family variably affected by BFRBDs and believe, to our knowledge, that this is the first report of 4 generations of a family in which individual members suffer from trichotillomania, skin-picking disorder, and nail biting with no other evidence of psychiatric disorders.

CASE REPORTS

Three members of a South African family present with a long-standing history of various BFRBDs.

The index case is a 41-year-old woman (III-5, Fig 1: pedigree) who presented with persistent acne and associated skin picking since the age of 15 years. The lesions are localized to the face with a mild premenstrual flare, with generally less than 5 lesions per cycle. Picked lesions become hyperpigmented (Fig 2) persisting for a variable time (weeks to months) after the patient moves onto new lesions. She had significant childhood nail biting, which resolved when the skin picking started. In addition, she suffers from allergic rhinitis requiring intermittent antihistamine treatment. Topical and systemic acne treatments were

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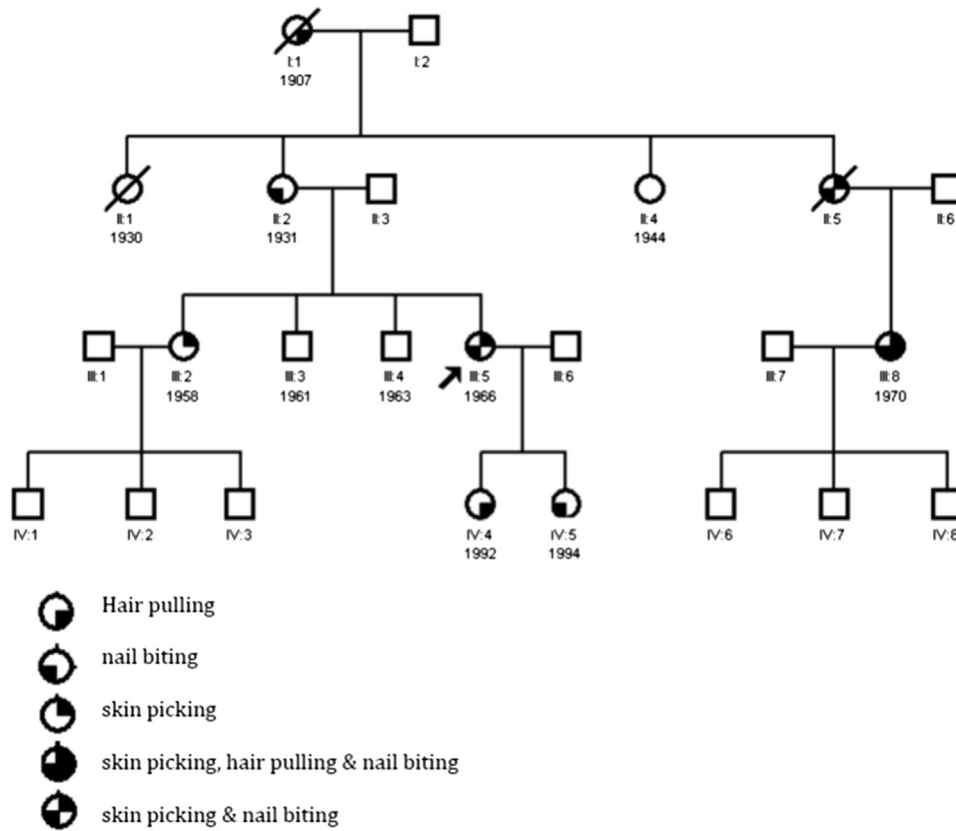


Fig 1. Family pedigree.



Fig 2. Case 1. Significant disfiguring hyperpigmentation of picked lesions.

interrupted over many years because of adverse effects aggravated by atopy; for example, topical tretinoin and doses of isotretinoin as low as 20 mg weekly resulted in excessively dry skin and worse picking.

The patient's 48-year-old sister (III-1) has a history of childhood asthma and teen-onset mild acne and severe persistent skin picking. The acne is mild but persists in spite of various treatments including courses of isotretinoin interrupted because of intolerable dryness. A first cousin (III-5) has similar mild acne but picks both the face and scalp with resultant patchy alopecia. The patient's mother (II-2) has atopy but no pathologic grooming. The maternal grandmother (I-1) until her death at age 98 years had

severe hair pulling and patchy hair loss but no atopy. The grandmother's pulling was so intense that a repetitive sound was audible when she ran her hair through her fingers. The patient has two daughters: one has asthma (IV-1) and hair pulling and the other nail biting (IV-2) but no atopy; both her brothers are unaffected, her sister and cousin each have 3 sons, none of whom are similarly affected (Fig 1).

DISCUSSION

Several clinical features are shared by the index case, her sister, and cousin, and are consistent with diagnostic criteria for trichotillomania and skin picking disorder in DSM-5. A central symptom is an uncontrollable urge to pick skin, an urge that is often triggered at times of decreased activity. It is accompanied by a thought that the roughness of the skin can be fixed by picking, by a feeling of relief at the time of picking, and by a strong sense of subsequent regret, particularly if picking leads to tenderness or bleeding. The picking is significant enough for other family members to notice it and discourage it. All three are successful professionals with no history of other psychiatric conditions.

These cases are consistent with the move in DSM-5 and International Classification of Diseases 11th

Revision to classify trichotillomania, skin-picking disorder, and other BFRBDs in the same section of the nomenclature.^{3,11} These cases are also consistent with more systematic studies of the familial relationships between different grooming disorders.⁸ In addition, they are consistent with previous reports emphasizing that BFRBDs have an early onset and are more common in women. This report adds to the literature by showing the possibility that BFRBDs can occur over 4 generations and in the absence of comorbidity with other psychiatric disorders. However, psychiatric disorders may have been missed because structured diagnostic interviews were not undertaken with family members.

The finding that serotonin reuptake inhibitors are more effective than noradrenergic reuptake inhibitors, not only in obsessive-compulsive disorder, but also in trichotillomania¹² and severe nail-biting,¹³ encouraged biological research on a range of putative obsessive-compulsive and related disorders. Nevertheless, much remains to be learned about the relevant psychobiology of the BFRBDs. Case reports such as these may also be useful in drawing attention to possibly overlooked factors. For example, comorbidity with atopy in our cases was inconsistent but may raise the question of whether certain underlying biological mechanisms are involved in both atopy and BFRBDs.

Future advances in understanding the familial and genetic risk factors for trichotillomania, skin-picking disorder, and other BFRBDs may help lead to novel treatments. In the interim, it is important for dermatologists to be aware of a number of key points about management. First, despite the high prevalence of these conditions, patients are often embarrassed by and ashamed of their symptoms so may not volunteer them unless specifically asked.⁵ Second, the increase of consumer advocacy groups has been important in addressing such issues; indeed, consumer support played a key role in the decision to include skin-picking disorder in DSM-5.¹⁴ Third, several efficacious pharmacotherapeutic and psychotherapeutic interventions are now available for the management of these conditions.^{15,16}

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