

University of Groningen

A case of follicular keratosis with trichostasis on an amputated limb

Meulenbelt, HEJ; Geertzen, JHB; Kardaun, SH; Jonkman, MF

Published in:
Clinical and Experimental Dermatology

DOI:
[10.1111/j.1365-2230.2006.02145.x](https://doi.org/10.1111/j.1365-2230.2006.02145.x)

IMPORTANT NOTE: You are advised to consult the publisher's version (publisher's PDF) if you wish to cite from it. Please check the document version below.

Document Version
Publisher's PDF, also known as Version of record

Publication date:
2006

[Link to publication in University of Groningen/UMCG research database](#)

Citation for published version (APA):

Meulenbelt, HEJ., Geertzen, JHB., Kardaun, SH., & Jonkman, MF. (2006). A case of follicular keratosis with trichostasis on an amputated limb. *Clinical and Experimental Dermatology*, 31(4), 600-601.
<https://doi.org/10.1111/j.1365-2230.2006.02145.x>

Copyright

Other than for strictly personal use, it is not permitted to download or to forward/distribute the text or part of it without the consent of the author(s) and/or copyright holder(s), unless the work is under an open content license (like Creative Commons).

The publication may also be distributed here under the terms of Article 25fa of the Dutch Copyright Act, indicated by the "Taverne" license. More information can be found on the University of Groningen website: <https://www.rug.nl/library/open-access/self-archiving-pure/taverne-amendment>.

Take-down policy

If you believe that this document breaches copyright please contact us providing details, and we will remove access to the work immediately and investigate your claim.

Downloaded from the University of Groningen/UMCG research database (Pure): <http://www.rug.nl/research/portal>. For technical reasons the number of authors shown on this cover page is limited to 10 maximum.

A case of follicular keratosis with trichostasis on an amputated limb

doi: 10.1111/j.1365-2230.2006.02145.x

A 29-year-old woman was referred to the Center for Rehabilitation, with complaints of pain and loss of mobility of the stump of her left leg. In 1999, she had undergone a transfemoral amputation because of complex regional pain syndrome (CRPS) type 1 of her left lower limb.

At the time of her presentation in 2003, she had no signs of CRPS type 1, as described by the International Association for the Study of Pain, on the stump.¹ She did not wear a prosthesis or clothing over the amputated side, because of persistent touch-evoked pain. She was not able to notice warm or cold sensations with the stump, and it is unknown for how long this had existed. She ambulated by using crutches and a wheelchair.

Physical examination showed an obese woman with a giant transfemoral stump with a surplus of soft tissue. The stump felt cold. The distal part of the stump showed a circumscribed asymptomatic area consisting of multiple hyperkeratotic follicular papules and hyperpigmentation (Fig. 1). This skin condition was not present prior to amputation. Potassium-hydroxide preparations and cultures for fungus and bacteria were negative. Vascular examination by Doppler ultrasound showed no major abnormalities in the blood flow of the stump. A skin biopsy taken from the affected area was not representative, showing only dermal oedema.

Physical examination 3 months later revealed erythema on the stump with a circumscribed field of large yellowish brown keratotic papules with follicular distribution (Fig. 1, inset). Histopathological examination of a second biopsy showed a very wide distended follicle containing a keratotic plug consisting of laminated and amorphous keratin containing several vellus hairs in a bundle (double refractile with polarization). The follicular wall matured normally. The histopathological diagnosis



Figure 1 Transfemoral stump with large circumscribed follicular keratotic papules. Inset: close-up view.

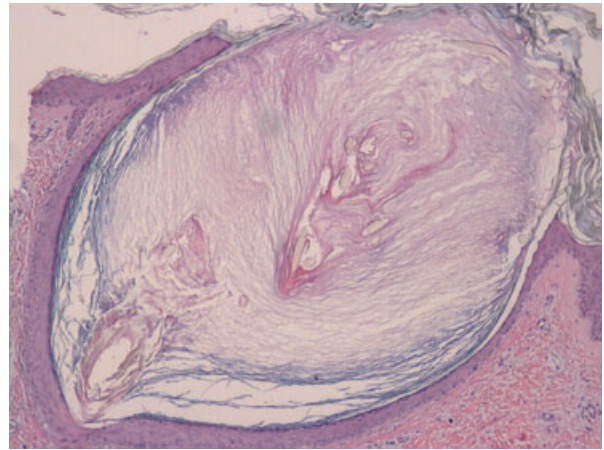


Figure 2 Skin histology shows a distended follicle with a horny plug with many cross-sections of vellus hairs. The superficial dermis shows a very mild perivascular lymphocytic inflammatory infiltrate (haematoxylin and eosin, original magnification $\times 4$).

was compatible with follicular keratosis with trichostasis (Fig. 2).

In order to enhance mobility, partial distal resection of the stump, including the affected skin, was performed. Over the following 6 months, the new stump with unaffected skin returned to the situation prior to surgery, i.e. follicular hyperkeratosis. Complaints of touch-evoked pain did not improve after resection, and mobility of the stump was not enhanced. A third biopsy of the affected area of the stump again showed a distended follicle containing several vellus hairs and follicular plugging, but also extrafollicular vellus hairs evoking a foreign-body cell reaction.

Follicular keratosis of amputated sites is not uncommon and is confined to sites with continuous friction caused by an ill-fitting prosthesis.^{2,3} Unlike the cases described by Ibbotson *et al.*,² our patient never used a prosthesis, owing to touch-evoked pain of the stump, thereby excluding friction as cause of the skin condition. The condition in our patient could be classified as follicular keratosis with secondary vellus hair retention caused by the hyperkeratotic plug. Retention of vellus hairs is named trichostasis. Trichostasis spinulosa is a not uncommon skin disorder that presents as follicular keratosis, but we prefer to use the diagnosis 'follicular keratosis with trichostasis' for our patient, as large yellowish-brown, asymptomatic, hyperkeratotic papules were observed, whereas in trichostasis, mildly pruritic and elevated, raspy, spinous, follicular plugs are most frequently seen.

Trichostasis results from successive production of vellus telogen club hairs from a single hair matrix in a follicle.⁴ There are several speculations regarding the aetiology of damage to hair follicles, such as external irritants or development of numerous resting buds in response to an unknown stimulus.⁵ There are two types of trichostasis spinulosa: the classical type with nonitching, solitary

comedo-like lesions on the face, especially on the nose in the elderly, and the itching type with multiple fine follicular papules, mainly located on the trunk and arms. The latter type may resemble keratosis pilaris at first sight.⁴

We do not know the mechanism that has led to the follicular keratosis, but we think that it caused secondary trapping of vellus hairs, which may have led to follicular rupture and subsequent foreign-body reaction to extra-follicular keratins in the dermis. It may be speculated that in our patient with CRPS type 1, the amputation in addition to an unknown factor could have led to an exaggeration of the normal cycle of the hair follicle or damage to the hair follicle, resulting in the development of numerous resting buds or prevention of the normal shedding of hair.

We conclude that the clinical presentation of follicular keratosis with secondary trichostasis on the stump of an amputee without prosthesis appears unique, and has to the best of our knowledge not been reported previously.

H. E. J. Meulenbelt,*‡ J. H. B. Geertzen,*‡ S. H. Kardaun† and M. F. Jonkman†

*Center for Rehabilitation, †Department of Dermatology, and

‡Northern Centre for Health Care Research, University Medical Center Groningen, University of Groningen, the Netherlands

E-mail: h.e.j.meulenbelt@rev.umcg.nl

Conflict of interest: none declared.

Accepted for publication 10 February 2006

References

- 1 Merskey H, Bogduk N. *Classification of Chronic Pain: Descriptions of Chronic Pain Syndromes and Definition of Terms*, 2nd edn. Seattle: IASP Press, 1994.
- 2 Larregue M, Babin P, Gallet P *et al*. Penetrating hyperkeratosis of an amputated limb. *Bull Soc Fr Dermatol Syphiligr* 1975; **82**: 462–4.
- 3 Ibbotson SH, Simpson NB, Fyfe NC *et al*. Follicular keratoses at amputation sites. *Br J Dermatol* 1994; **130**: 770–2.
- 4 Strobos MA, Jonkman MF. Trichostasis spinulosa: itchy follicular papules in young adults. *Int J Dermatol* 2002; **41**: 643–6.
- 5 Young MC, Jorizzo JL, Sanchez RL *et al*. Trichostasis spinulosa. *Int J Dermatol* 1985; **24**: 575–80.

Pustular psoriasis over keloids: is it a Koebner phenomenon?

doi: 10.1111/j.1365-2230.2006.02159.x

A 44-year-old woman, who was known to have acrodermatitis continua of Hallopeau, presented with a flare of her disease over her hands and a 2-month history of development of pustular lesions over keloids located on her pre-sternal area. Her disease had started 2 years previously on

the left fourth finger, and had gradually progressed to involve other fingers. Keloids on her chest had arisen spontaneously without any preceding trauma 5 years previously. She was being managed with topical steroids and dapsone (100 mg oral tablet) daily, with partial response.

On examination, there were erythematous scaly plaques studded with pustules on the fingers, and there was tapering of the fingers. Some of her fingernails had fallen off following the coalescence of pustules and formation of lakes of pus on the nail bed. Besides the pustules over the keloids, a few pustules and some erythema were seen at the periphery of the keloids (Fig. 1). No other site besides the hands and chest were involved. Histopathology of the pustular lesions over keloids was consistent with pustular psoriasis (Fig. 2).

In 1878, Heinrich Koebner first described the development of an overt lesion of psoriasis in the traumatized uninvolved skin of patients. He called it the Koebner or isomorphic phenomenon.¹ His initial observations and studies resulted from having seen patients who had developed psoriasis at sites of excoriations, horse bites, and tattoos.

The Koebner phenomenon has been reported in a number of cutaneous diseases, and is frequent in psoriasis.² The incidence of a positive Koebner phenomenon in psoriasis is reported as 24–54% in published studies. The latent period between injury to uninvolved skin and appearance of disease is usually 10–14 days, but it may range from 3 days to several years. The Koebner response in psoriasis patients is noted significantly more frequently in patients with disease onset at a young age, in severe forms of disease, when the disease is unstable or flaring, and when the disease is resistant to therapy.³

The pathogenesis of Koebner phenomenon is not known. Speculative pathogenetic factors involved are immunological, vascular, dermal, enzymatic, inhibitory, neural, growth, genetic and hormonal. There is growing evidence that immunological factors are involved in the pathogen-



Figure 1 Pustular lesions over keloids on the chest.