

Acantholytic acanthoma on the skin of an amputated limb stump *Amputasyon güdüğünde gelişmiş akantolitik akantoma*

Banu Lebe¹, H. Uğur Pabuççuoğlu¹,

¹ Department of Pathology, Dokuz Eylül University School of Medicine, Inciraltı, İZMİR

We herein report a rare case of acantholytic acanthoma in a 42-year-old male patient. The tumour was located on the skin of the amputated right lower limb stump and the patient was using prosthesis. The physical effects of the prosthesis may be considered in the histogenesis of this entity, although convincing data does not exist. To the best of our knowledge acantholytic acanthoma in this localization has not been previously reported.

Key Words: Acantholytic acanthoma, amputation stump.

Burada 42 yaşında erkek hastada seyrek görülen akantolitik akantom olgusu sunulmuştur. Hasta protez kullanmaktadır ve tümör, ampute edilmiş sağ bacak güdüğü derisi üzerinde lokalizedir. Bu antitenin histogenezinde protezin fiziksel etkileri dikkate alınabilir, ancak inandırıcı bilgiler bulunmamaktadır. Bilgilerimize göre bu lokalizasyonda akantolitik akantom olgusu bildirilmemiştir.

Anahtar Kelimeler: Akantolitik akantom, amputasyon güdüğü.

Acanthoma is a benign tumour of keratinocytes. It can be classified as acantholytic, epidermolytic, clear cell, large cell acanthomas and acanthoma fissuratum^(1,2). Acantholytic acanthoma is typically asymptomatic, solitary, hyperkeratotic papule or nodule located predominantly over the trunk and occasionally extremities, histologically showing hyperkeratosis, papillomatosis, acanthosis and acantholysis. Acantholysis is the most prominent feature occurring in various levels of epidermis which may be combined with dyskeratosis^(1, 3-5).

We herein report a case of acantholytic acanthoma occurring on the skin of the amputated right lower limb stump of a 42-year-old male patient who was using bilateral prosthetic limbs subsequent to bilateral above knee amputation which was performed 35 years ago.

The purpose of this paper is to report an additional case with an unusual location and briefly review the histopathological differential diagnosis of this rare type of acanthoma. To the best of our knowledge, acantholytic acanthoma on the skin of the amputation stump has not been previously reported in the English language literature.

CASE REPORT

A 42-year-old man was admitted for the pruritic lesion on the skin of amputated right lower limb stump. The patient had a history of bilateral above knee amputation and subsequent right hand amputation performed 35 years ago. Medical records were not available but patient's history were compatible with a probable vascular disorder eventually leading gangrene. He had been using bilateral prosthetic limbs. He had also been operated for the squamous cell carcinoma of the right temporal skin 2 years ago, and at present he has no evidence of disease.

Clinical examination demonstrated a crusty, keratotic lesion. Clinical diagnosis of squamous cell carcinoma or dermatitis was made and an excisional biopsy was performed. The tissue received for histopathological examination was 35x10x5 mm. in dimensions with an irregular surface. All of the tissue was routinely processed, embedded in paraffin and 5 µm thick sections were stained with H&E.

Histopathological examination revealed a neoplastic keratinocytic lesion exhibiting hyperkeratosis and parakeratosis, irregular acanthosis, cleft like spaces and marked acantholysis at all levels with accompanying dyskeratotic cells (Figs. 1, 2, 3). These dyskeratotic cells were characterized by dense eosinophilic cytoplasm with well demarcated cytoplasmic borders. Mild perivascular lymphocytic infiltrate was present in the papillary dermis. The histopathological diagnosis was acantholytic acanthoma. The patient has been in good health without recurrence after 7 months.



Figure 1: Acantholytic acanthoma. Acantholytic spaces throughout epidermis with overlying prominent hyper-parakeratosis (HE, scanning magnification, x20).



Figure 2: Acantholytic acanthoma. Irregular acantholytic spaces with dyskeratotic cells (arrow) (HE, x200).

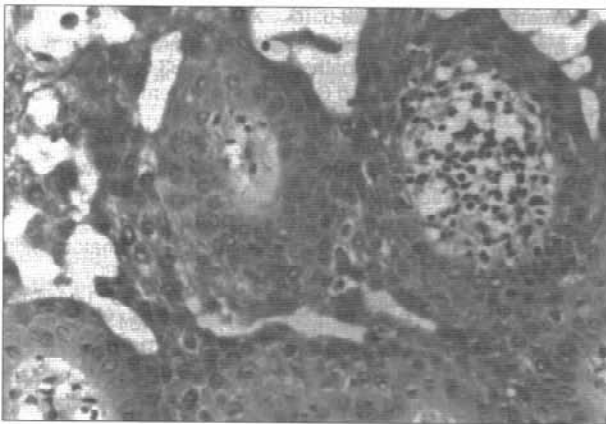


Figure 3: Higher magnification of cleft like and irregular spaces with acantholytic cells (H&E, x200).

DISCUSSION

Acantholytic acanthoma, was first described by Brownstein in 1988⁽³⁾ as a solitary benign cutaneous tumour presenting clinically as an asymptomatic keratotic papule or nodule. In this original report, which included 31 examples, the tumour was twice as common in males as females and half of the cases were on the trunk. Histologically the lesions were characterized by hyperkeratosis, papillomatosis, acanthosis and acantholysis. In this type of acanthoma, acantholysis was the outstanding histopathological finding. Mehaged et al⁽⁴⁾ reported a case of acantholytic acanthoma in a 61-year-

old man. The tumour was localized on the arm and showed parakeratosis in vertical orientation in addition to hyperkeratosis and irregular epithelial hyperplasia with acantholytic suprabasal clefts. Ramos-Caro et al⁽⁶⁾ reported multiple acantholytic acanthomas on the right arm of a man who underwent a renal transplantation 13 years ago. These lesions showed suprabasal acantholysis and focal mild dyskeratosis.

Acantholytic dyskeratotic acanthoma is a keratosis usually presenting as a solitary lesion⁽⁷⁾. When acantholysis is combined with dyskeratosis the tumour histologically resembles Darier's disease⁽⁸⁾. The wide list of diverse disorders that should be included in the histopathological differential diagnosis of acantholytic acanthoma may be summarized as follows: acantholytic seborrheic keratosis, acantholytic solar keratosis, warty dyskeratoma, localized Darier's disease, acantholytic dyskeratotic epidermal nevus, Hailey Hailey disease, incidental focal acantholytic dyskeratosis and immunologic disorders such as pemphigus vulgaris and pemphigus vegetans^(3, 4, 7, 8-12).

Acantholytic seborrheic keratosis differs from the acantholytic acanthoma by showing limited acantholytic change and having a classical architectural configuration with well known horn cysts^(1, 4). In acantholytic solar keratosis keratinocytic atypia is the hallmark and acantholysis occurs above the atypical cells of the basal layer⁽⁴⁾. Warty dyskeratoma usually presents as a solitary nodule and microscopically displays a cup-shaped invagination of a cystically dilated hair follicle with a keratin plug. Presence of Darier-like clefts containing acantholytic and dyskeratotic cells may lead to a confusion

with acantholytic dyskeratotic acanthoma. But the absence of the central cup-shaped cystic cavity with papillary projections and the lack of keratin plug in acantholytic acanthoma help to exclude the possibility of warty dyskeratoma ^(1, 4, 6).

It may be impossible to distinguish acantholytic dyskeratotic acanthoma from a solitary papule of Darier's disease in small biopsy specimens ⁽⁷⁾. Darier's disease is characterized by papillomatosis, acanthosis, hyperkeratosis, dyskeratosis and suprabasal acantholysis. Acantholytic dyskeratotic epidermal nevus also shows histopathological features similar to those of Darier's disease ^(7, 10, 11). It has been suggested that at least some of the reported acantholytic dyskeratotic epidermal nevi represented mosaic forms of Darier's disease ⁽⁹⁾ and evidence existed for classification some of these cases as localized Darier's disease ⁽⁸⁾. Focal acantholytic dyskeratosis has features in common with Darier's disease and acantholytic acanthoma such as suprabasal acantholysis and dyskeratosis, and these changes have also been encountered as an incidental finding in a variety of skin lesions including inflammatory and neoplastic conditions ⁽¹²⁾. The lesions of Hailey Hailey disease, pemphigus vulgaris and vegetans may mimic acantholytic acanthoma. However, clinical picture in these acantholytic disorders is entirely different than acantholytic acanthoma. It seems to us, in the histopathological differential diagnosis of acantholytic dyskeratotic acanthoma from Darier's disease and from the disorders with changes just like those of Darier's disease, clinico-pathological correlation may be mandatory for accurate diagnosis.

Roten and Bhawan ⁽²⁾ described a solitary hyperkeratotic lesion with epidermal changes characterized by epidermal hyperplasia, parakeratosis and presence of dyskeratotic cells at all levels of acanthotic epidermis. They proposed the term "dyskeratotic acanthoma" for this lesion. Although acantholysis is absent in this type of acanthoma, it may be included in the differential diagnosis.

To the best of our knowledge, occurrence of acantholytic acanthoma on the skin of the amputation stump has not been reported before. Skin problems affecting the limb stump of amputees or the prosthetic limb users are not uncommon. In a recent review of 210 cases with prosthetic limb, Lyon et al ⁽¹³⁾ observed a total of 71 patients with skin problems. Sixty of these patients had lower limb amputations. In 63 of these 71 patients the authors described a variety of dermatoses including physical dermatoses such as follicular hyperkeratosis, verrucous hyperplasia and epidermoid cysts. The etiology of acantholytic dyskeratotic acanthoma is unknown

^(4, 6). In an above mentioned report ⁽⁶⁾ this lesion was encountered in a immunosuppressed patient. That patient also had multiple basal and squamous cell carcinomas of the skin. Although our patient had been operated for the squamous cell carcinoma of the temporal skin, he had no evidence of immunosuppression. In our case, it may be speculated that acantholytic acanthoma could be related to localized trauma, due to physical effects of the prosthesis. But, convincing evidence does not exist.

In summary we have described a solitary acquired lesion characterized by hyperkeratosis, parakeratosis, acanthosis, acantholysis and dyskeratosis representing acantholytic dyskeratotic acanthoma. The tumour was localized on the skin of an amputation stump and the patient had no previous history of immunologic or non-immunologic acantholytic disorders and no evidence of immunosuppression.

REFERENCES

1. Brownstein MH. The benign acanthomas. *J Cutan Pathol* 1985; 12: 172-88.
2. Roten SV, Bhawan J. Isolated dyskeratotic acanthoma. A variant of isolated epidermolytic acanthoma. *Am J Dermatopathol* 1995; 17: 63-6.
3. Brownstein MH. Acantholytic acanthoma. *J Am Acad Dermatol* 1988; 19: 783-6.
4. Megahed M, Kochanek-Scharffetter K. Acantholytic acanthoma. *Am J Dermatopathol* 1993; 15: 283-5.
5. Kirkham N. Tumors and Cysts of the Epidermis. In: Elder D, ed. *Lever's Histopathology of the Skin*. Philadelphia: Lippincott-Raven Publishers. 1997: 687-8.
6. Ramos-Caro F, Sexton M, Browder JF, Flowers FP. Acantholytic acanthomas in an immunosuppressed patient. *J Am Acad Dermatol* 1992; 27: 452-3.
7. Ackerman AB. *Histologic Diagnosis of Inflammatory Skin Diseases*. Baltimore: Williams & Wilkins. 1997: 807-8.
8. O'Malley MP, Haake A, Goldsmith L, Berg D. Localized Darier disease. *Arch Dermatol* 1997; 133: 1134-8.
9. Cambiaghi S, Brusasco A, Grimalt R, Caputo R. Acantholytic dyskeratotic epidermal nevus as a mosaic form of Darier's disease. *J Am Acad Dermatol* 1995; 32: 284-5.
10. Cottoni F, Masala MV, Cossu S. Acantholytic dyskeratotic epidermal nevus localized unilaterally in the cutaneous and genital areas. *Br J Dermatol* 1998; 138: 875-8.
11. Youn M, Hann SK, Moon TK, Lee MG. Acantholytic dyskeratotic epidermal nevus induced by ultraviolet B radiation. *J Am Acad Dermatol* 1998; 39: 301-4.
12. DiMaio DJM, Cohen PR. Incidental focal acantholytic dyskeratosis. *J Am Acad Dermatol* 1998; 38: 243-7.
13. Lyon CC, Kulkarni J, Zimerson E, Van Ross E, Beck ME. Skin disorders in amputees. *J Am Acad Dermatol* 2000; 42: 501-7.