

# Multiple acantholytic dyskeratotic acanthomas in an organ transplant recipient



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**Key words:** heart transplantation; immunosuppression; multiple acantholytic dyskeratotic acanthomas.

## INTRODUCTION

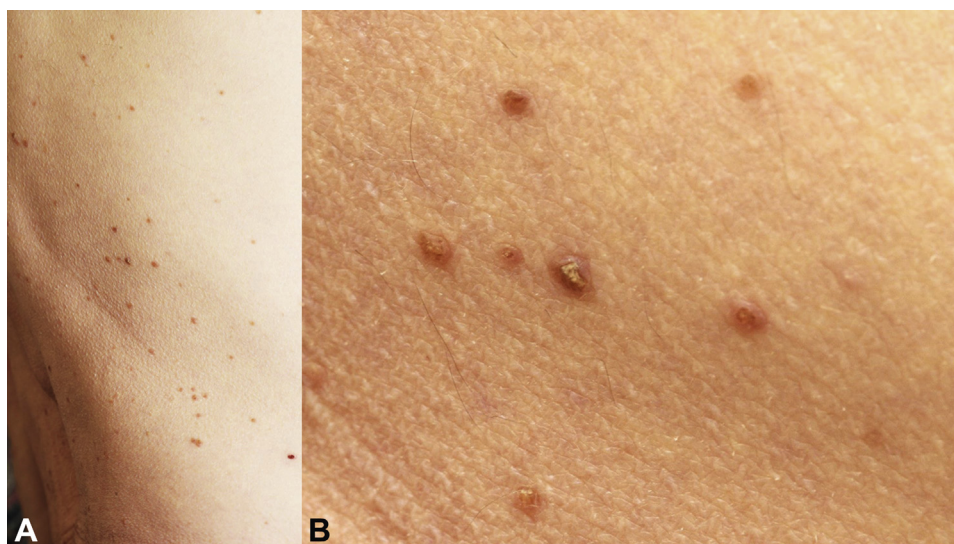
Organ transplant recipients are known for carrying a high risk of various benign and malignant tumor development, particularly of the skin. Therapeutic immunosuppression in association with a variety of (co-)carcinogenic factors are held responsible for the high tumor incidence.

Acantholytic dyskeratotic acanthoma is a rare benign condition characterized by circumscribed

epidermal proliferation displaying both acantholysis and dyskeratosis. Clinically, most cases were first diagnosed as basal cell carcinoma, which prompted their excision and histopathologic examination.

## CASE REPORT

A 60-year-old man underwent heart transplantation because of severe dilatative cardiomyopathy in 2016. He received mycophenolate mofetil, tacrolimus,



**Fig 1.** Acantholytic dyskeratotic acanthomas. Clinical aspect of the lesions on the lower back. **A**, Overview shows the distribution of the lesions. **B**, Detailed clinical aspect of a lesion on the back.

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Funding sources: None.

Conflicts of interest: None disclosed.

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JAAD Case Reports 2018;4:695-7.  
 2352-5126

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<https://doi.org/10.1016/j.jidcr.2018.04.009>

everolimus, and prednisone for prevention of organ rejection.

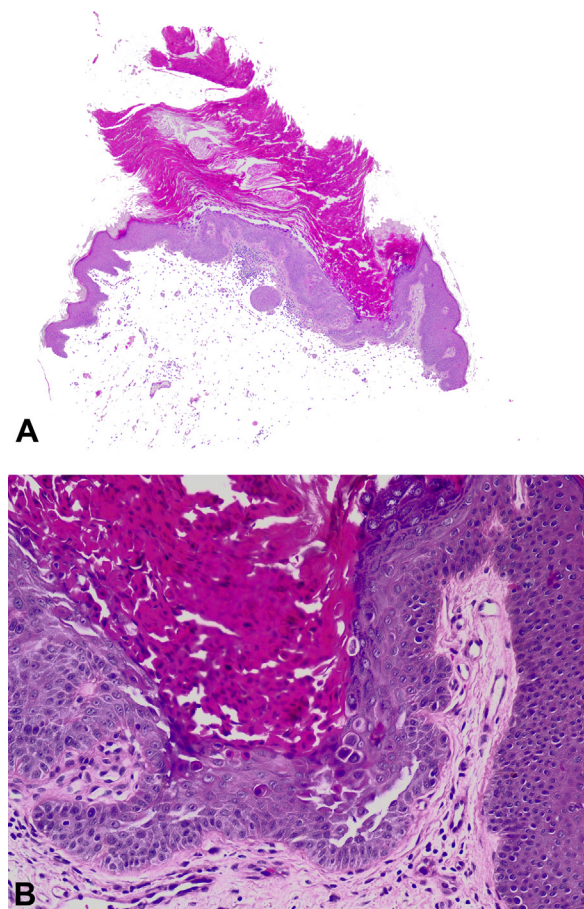
In October 2017, a week after his wife had noticed the lesions, he presented with multiple asymptomatic disseminated papules on his back and lateral chest wall that demonstrated a slight central umbilication or core (Fig 1). The patient had not received dermatologic care before. No skin malignancies were detected on full-body examination. The nails were completely normal. Clinically, a perforating collagenosis was suspected. A biopsy section was taken for histopathologic examination.

The sections were stained with hematoxylin-eosin and periodic acid–Schiff. Serial and step sections showed very characteristic changes. On one side of the lesion, there was an angled indentation of the epidermis with marked dyskeratosis and acantholysis of the keratinocytes in the bottom (Fig 2, A). In particular, some very large acantholytic dyskeratotic cells with hyperchromatic nuclei and eosinophilic perinuclear shells representing rings of condensed tonofilaments around the nucleus were observed (Fig 2, B). Otherwise there was no conspicuous cellular atypia. From here, a column of dyskeratotic cells extended obliquely upward, giving the lesion a slight resemblance to porokeratosis. In the first biopsy, the acantholysis extended to the side, to which the dyskeratotic column was leaning, but it became more superficial and almost no dyskeratoses were formed here. Based on these observations, an acantholytic dyskeratotic acanthoma was diagnosed.

Because of this unique finding, 2 more excisions were performed that showed the same histopathologic changes, thus confirming that all lesions were indeed acantholytic dyskeratotic acanthomas.

## DISCUSSION

Acantholytic dyskeratotic acanthoma is a rare benign lesion of unknown etiology and pathogenesis.<sup>1</sup> The striking features of acantholysis and dyskeratosis are also seen in other dermatoses,<sup>2</sup> particularly Darier dyskeratosis follicularis, pemphigus benignus familiaris of Hailey-Hailey, Grover transitory acantholytic dermatosis, warty dyskeratoma,<sup>3</sup> acantholytic dyskeratotic epidermal nevus,<sup>4</sup> and acantholytic squamous cell carcinoma.<sup>5</sup> Rarely, acantholytic acanthomas without major dyskeratosis or dyskeratotic acanthomas without acantholysis are seen.<sup>6,7</sup> However, dyskeratotic acanthoma is not a variant of epidermolytic acanthoma,<sup>2,8</sup> as dyskeratosis and epidermolytic hyperkeratosis with its underlying granular degeneration are fundamentally different processes. Most acantholytic dyskeratotic acanthoma lesions are localized



**Fig 2.** Histopathology of an acantholytic dyskeratotic acanthoma. **A**, Scanning magnification of the tumor shows a hyperkeratotic acanthoma with indentation on one side, an obliquely upward-pointing column of dyskeratotic cells, and marked acantholysis over most of the tumor width. **B**, Higher magnification shows the suprabasal acantholysis, dyskeratoses, and abnormally large round to oval cells with a clear cytoplasmic periphery. (**A** and **B**, Hematoxylin-eosin stain; original magnifications: **A**,  $\times 40$ ; **B**,  $\times 200$ .)

on the trunk.<sup>9</sup> Subungual localization was published in 3 cases<sup>10</sup> and seen by us in 4 cases (unpublished).

As the main differential diagnosis in our case, we ruled out Grover transitory acantholytic dermatosis because of clinical features of missing pruritus and distribution (disseminated lesions just on the back) and histologic features; an asymmetric angled indentation, such a high amount of dyskeratotic hyperkeratosis, and the column of dyskeratotic cells are not seen in Grover transitory acantholytic dermatosis. We also ruled out multiple warty dyskeratomas because this entity histologically shows a symmetric endophytic tumor, which is not the case in our patient. None of the other above-mentioned dermatoses with acantholysis and dyskeratosis are clinically or histologically consistent with our case.

The etiopathogenesis of acantholytic dyskeratotic acanthoma remains unknown. One case was described in a patient undergoing vemurafenib therapy for metastasizing melanoma, and a mechanism similar to that causing multiple keratoacanthomas in these patients was suggested.<sup>8</sup> We are not aware that any of the immunosuppressive drugs taken by our patient has a similar action. However, immunosuppression might allow cell clones prone to undergo dyskeratosis and acantholysis to proliferate, which otherwise might have been eliminated by the immune system.

To the best of our knowledge, multiple acantholytic dyskeratotic acanthomas have not yet been described. It is tempting to speculate that the immunosuppression necessary in organ transplant recipients may have prevented the elimination of those cell clones that give rise to this peculiar benign lesion.

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