

## A Case of Focal Acantholytic Dyskeratosis Occurring on both the Lip and the Anal Canal

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Focal acantholytic dyskeratosis has a distinctive histological pattern that is associated with various clinical expressions. It rarely occurs on the lip or the perianal area. We report a patient with focal acantholytic dyskeratosis occurring on both the upper lip and the anal canal. Histopathologically, the lesions showed hyperkeratosis, suprabasilar clefting, epidermal acantholysis and dyskeratosis. This case represents the first report of a focal acantholytic dyskeratosis occurring on both the lip and the anal canal.

**Key Words:** Focal acantholytic dyskeratosis, lip, anal canal

### INTRODUCTION

Focal acantholytic dyskeratosis (FAD) is a rare condition that is characterized by the distinct histological patterns of suprabasilar cleft, acantholysis, dyskeratosis, hyperkeratosis, parakeratosis, and varying degrees of basal cell hyperplasia.<sup>1</sup>

Several cases of FAD of the lip,<sup>2,4</sup> genital<sup>5-8</sup> or perianal region<sup>9,10</sup> have been reported, but there have been no reports of FAD occurring both on the lip and the perianal regions. We report a case of FAD in a 41-year-old man occurring on both the upper lip and the anal canal.

### CASE REPORT

A 41-year-old man visited our clinic with wet

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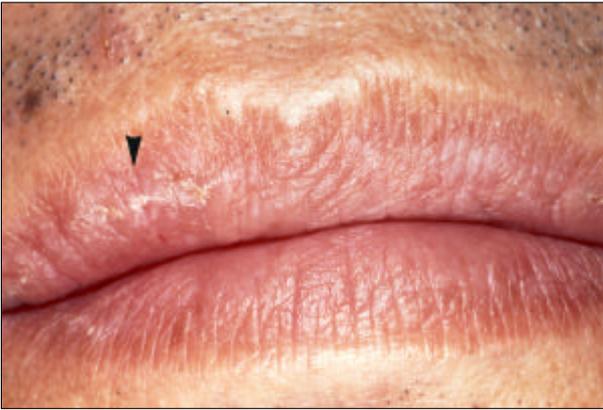
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swollen lesions on the lip and the anal canal that first occurred approximately 8 months prior. A physical examination revealed multiple 2-3 mm sized slightly hydrated crusted papules and confluent patches on the upper lip (Fig. 1), with multiple variable sized whitish hydrated papules on the anal canal. There were no other specific skin lesions. He had undergone a hemorrhoidectomy two years ago. The laboratory findings including a complete blood count, liver and renal function tests, and urinalysis were within the normal limits. Skin biopsies from the upper lip and the anal canal showed hyperkeratosis, parakeratosis, acanthosis, suprabasilar and intra-epidermal clefting throughout the entire thickness of the epidermis, and acantholytic cells were found overlaying the villi (Fig. 2A and 2B). Some dyskeratotic cells with eosinophilic cytoplasm and small, darkly-staining nuclei were observed above the clefts in the lip specimen. However, dyskeratosis was rarely observed in the anal specimen.

### DISCUSSION

FAD, which was initially described by Ackerman,<sup>1</sup> has a histological pattern that is characterized by suprabasilar clefts, acantholytic and dyskeratotic cells, and hyperkeratosis. Acantholytic dyskeratosis is the main histological feature of various skin diseases including Darier's disease, Grover's disease, Hailey-Hailey disease, and warty dyskeratoma. These diseases could be excluded in this case. FAD may also be observed focally in association with basal cell epithelioma,

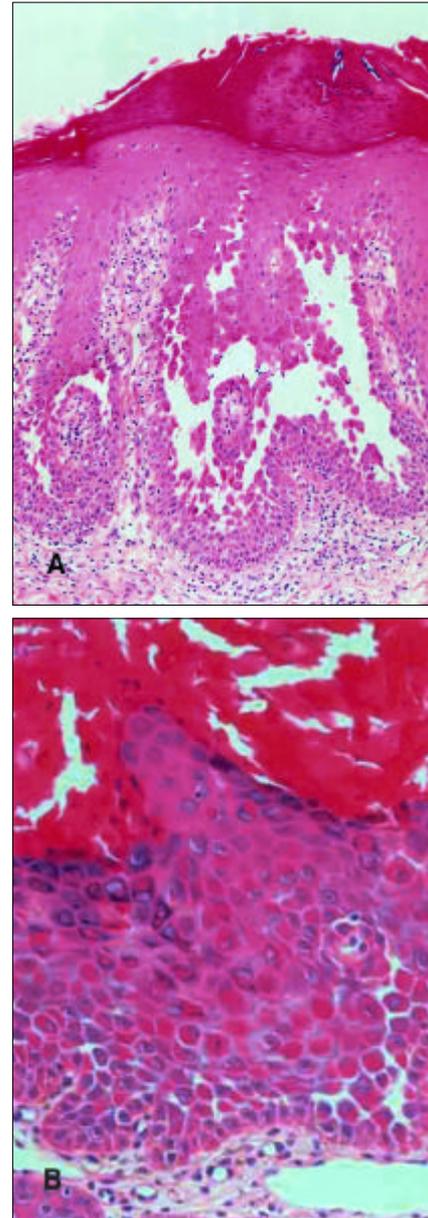


**Fig. 1.** Multiple ill-defined 2-3 mm sized hydrated crusted papules are presented on the upper lip.

dermatofibroma, chondrodermatitis nodularis helices and comedones.<sup>1</sup> However, in our case, there were no associated skin diseases.

The term 'papular acantholytic dyskeratosis' has been applied to FAD with multiple lesions developing on the vulva, the perianal area, or the penis, and the term 'papular acantholytic dyskeratoma'<sup>4</sup> has been applied to clinically apparent solitary lesions. The former is also classified as acantholytic dermatosis of the genitocrural/perianal region.<sup>6,8</sup> Papular acantholytic dyskeratosis usually exhibits prominent acantholysis with little or no dyskeratosis,<sup>6</sup> which is consistent with the histopathological findings of the lesion on the anal canal in this case. Lip or anal involvements of FAD are rare, among which only 4 cases on the lip<sup>2-4</sup> and 2 cases on the anal canal<sup>9,10</sup> have been reported. To our knowledge, this is the first case of FAD on both the lip and the anal canal.

As the etiology and pathogenesis of FAD, trauma and tobacco use as a source of chronic irritations have been suggested.<sup>3,4,9,10</sup> In previously reported cases on the lips, there was a history of tobacco use in 2 cases (50%). This patient also had a smoking history. Interestingly, there was a past history of hemorrhoidectomy in all 3 FAD cases (100%) on the anal canal including this case. The previous FAD cases in the anal canal documented an incidental form of FAD that was clinically unapparent. However, this patient showed clinically localized papular lesions on the anal canal. Furthermore, he also had similar lesions on the lip. Therefore, it is suggested that FAD on the lip



**Fig. 2.** (A) Hyperkeratosis, parakeratosis, acantholysis with suprabasilar clefting are observed in the lesion from the anus. Hematoxylin-eosin stain. Original magnification  $\times 100$ . (B) Numerous acantholytic cells and some dyskeratotic cells are present in the epidermis in the biopsy specimen from the lip. Hematoxylin-eosin stain. Original magnification  $\times 200$ .

and/or the anal canal, including this case, represents a localized form of FAD, and not an incidental form. It is proposed that chronic irritations to the mucosa like tobacco use and a hemorrhoidectomy may be associated with the etiology of FAD involving the mucosa. The identification

of FAD both on the lip and the anal canal suggests that FAD of the lip and that of the anal canal ought not be considered as separate entities and should be included in the same spectrum.

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