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Delayed Diagnosis of Squamous Cell Carcinoma of the Scrotum in a Patient with Behçet's Disease

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Dear Editor:

Behçet's disease (BD), a chronic inflammatory disease, is characterized by oral aphthae and genital ulcer, arthritis, cutaneous lesions, such as non-bacterial folliculitis, and erythema nodosum, as well as ocular, gastrointestinal and neurological manifestations. Although genital ulcers are histologically similar to oral aphthae, they are deeper than oral aphthous ulcers and are a major criterion for diagnosis. We present a case of scrotal squamous cell carcinoma (SCC) in a patient diagnosed with BD.

A 39-year-old male patient was admitted to our clinic with recurrent oral aphthae and a deep ulcer, covering the scrotum and perineal regions. The patient had been suffering from recurrent oral aphthous lesions for almost 11 years, and he had been medicated with 1.5 mg/day of colchicine, with a diagnosis of BD. Over the preceding 2 years, the genital ulcer had gradually enlarged and deepened. During this period, the patient received topical treatment and systemic antibiotics therapy; however, he was lost to follow-up.

Dermatological examination revealed a large and painless ulcer, extending from the scrotum to perineal region, which was approximately 8 cm in diameter (Fig. 1). This

ulcer had an indurated floor and also sharp and irregular edges. No growth was observed in the bacterial and fungal cultures of the genital ulcer. No inguinal lymphadenopathy was noted and syphilis tests were negative.

The ulcer had a foul odored heavy discharge; it was considered as a secondary bacterial infection and systemic antibiotics therapy was initiated. After the treatment of wound infection, the lesion was excised with a margin of at least 1 cm of normal skin. The surgical specimen showed neoplastic proliferation of squamous differentiated tumour cells in the epidermis (Fig. 2). The test of the tumor cells for anti-human papilloma viruses (HPVs) antibodies was negative; furthermore, HPVs DNA was not detected by direct polymerase chain reaction (PCR) method in excisional tissue specimen. In addition, abdominal, chest and pelvic computed tomography scan showed no evident distant metastasis, including the regional lymph nodes. Six months after surgical treatment, local recurrence or metastasis was not detected yet.

Genital ulcers in BD are sometimes large and deeply located; however, they heal with scarring in 4 weeks¹. Our patient had been suffering from genital ulcer for two

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Fig. 1. Well-demarcated large destructive ulcers on the scrotum and perineal area.

years. The ulcer had a nodulo-ulcerative appearance and its base was highly indurated. The clinical features of the ulcer and its unresponsiveness to treatment led us to investigate other possible causes of genital ulcer. We doubted whether the SCC developed on the chronic genital ulcer of BD or emerged directly as SCC, unrelated with genital ulcer. In fact, SCC was diagnosed at first by us during his first appointment at our clinic. However, we suggest that the lesion occurred directly as SCC, due to infiltration of the whole ulcer region and the rapid progression of the lesion.

Skin tumours in BD are rarely observed². *In situ* SCC development on a chronic vulvar ulcer of a female patient with BD was previously reported. The authors had pointed out that long-term systemic corticosteroid treatment could contribute to the development of SCC in this genital ulcer, which was resistant to treatment³. Satolli et al.⁴ reported a Merkel cell carcinoma in a patient with BD, and mentioned that the immunosuppressives may lead to the development of cancer. In our case, there was no predisposition for the development of SCC, such as

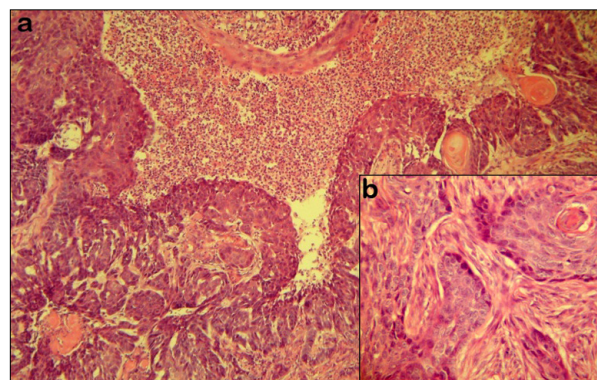


Fig. 2. (a) Histological picture of squamous cell carcinoma: Neoplastic proliferation of squamous differentiated tumour cells with dyskeratotic and atypical keratinocytes in the epidermis (H&E, $\times 200$). (b) Moderately differentiated tumour cells fill and expand papillary and reticular dermis (H&E, $\times 400$).

immunosuppressive drugs, arsenic exposure, irradiation, burn scarring, and HPV infection. We suppose that the presence of SCC together with BD is coincidental. Like BD, the other dermatoses, composed of various components, should be followed carefully, and differential diagnoses of signs and symptoms should be made. Especially, all clinical features of a genital ulcer should be noted and unusual causes should not be forgotten.

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