

A diagnostic challenge: chronic cutaneous ulcers as an isolated clinical finding in metastatic Crohn's disease

Keywords: Metastatic Crohn's disease. Disfiguring ulcers. Non-caseating granulomas.

Dear Editor,

Metastatic Crohn's disease (MCD) is the most uncommon cutaneous manifestation of Crohn's disease. The actual incidence is not clear and there are fewer than 200 cases described in the literature. MCD can appear in the absence of gastrointestinal involvement or precede it by several months or even years, especially during the pediatric age (1).

Case report

We report the case of a 21-year-old female that presented to our Emergency Care with large exudative ulcers in the inguinal folds, vulva, perianal region and popliteal fossae (Fig. 1A-C). Histopathological examination of the ulcers revealed a non-caseating granulomatous inflammation with abundant multinucleated giant cells and intense lymphocytic infiltrate (Fig. 1D). Colonoscopy, contrast study of the small bowel and video capsule endoscopy were carried out without evidence of digestive disease. High potency topical steroids (betamethasone 0.5 mg/g twice daily) and 300 mg intravenous infliximab were initiated as the initial line therapy, and the ulcers began to heal (Fig. 1E and F).

Discussion

MCD is a rare cutaneous condition with diverse locations, often arising at the perineum or skinfolds. Differential diagnoses include hidradenitis suppurativa, cutaneous sarcoidosis or tuberculosis, Crohn's disease and sexual abuse (2,3). In our case, the likelihood of an extraintestinal manifestation of Crohn's disease was not initially considered due to the absence of gastrointestinal complaints and rectal lesions. A detailed history and physical exam are needed, but a biopsy demonstrating non-caseating, epithelioid cell granulomas with a surrounding lymphoid infiltrate is mandatory for the diagnosis.

The treatment requires an interdisciplinary approach (2). No relationship has been demonstrated between MCD and the activity of the intestinal disease. In the majority of cases, cutaneous manifestations developed closely to a well-defined intestinal disease (3). However, perianal lesions arise before the development of the gastrointestinal symptoms in 20-30 % of patients (1). We report this case to highlight the presence

of cutaneous ulcers without intestinal disease in a young non-pediatric female as a unique manifestation of the disease.

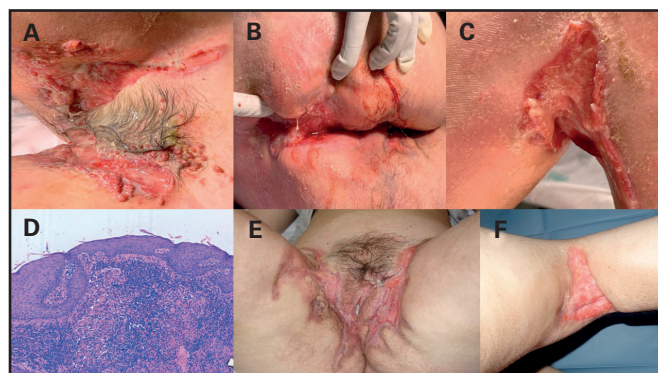


Fig. 1. Disfiguring ulcers affecting the vulva and the inguinal creases in a linear arrangement, with granulation tissue and polypoid papules at the periphery (A). Similar ulcers affecting the natal cleft (B) and both popliteal fossae (C). The presence of severe non-caseating granulomatous inflammation with abundant multinucleated giant cells and intense lymphocytic infiltrate. Epidermic spongiosis with lymphocytic exocytosis. No microorganisms were identified (D). Results after three weeks of treatment with topical betamethasone 0.5 mg/g twice daily and two doses of infliximab 300 mg: clear improvement of the lesions with scarring at the edges and some skin tags in the right thigh (E). Favorable evolution of the perianal region and the popliteal fossae (F).

Conflict of interest: the authors declare no conflict of interest.

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