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SHORT COMMUNICATION

Coexistence of Eccrine Squamous Syringometaplasia and Graft-Versus-Host Disease in a Patient with Secondary Myelodysplastic Syndrome

Raquel Carrascosa[1], Maria J. Concha[1], Javier Sánchez-Pérez[1], Maximiliano Aragües[1] and Javier Fraga[2]
Departments of [1]Dermatology and [2]Pathology, Hospital Universitario La Princesa, Diego de León, 62, ES-28006 Madrid, Spain. E-mail: rachel170786@gmail.com
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Eccrine squamous syringometaplasia (ESS) is defined as the metaplasia of eccrine duct cuboid epithelial cells in squamous cells (1). It was first described by Cornil in 1865 in the periphery of what was then called adenoacanthoma, which is today known as acantholytic, adenoid or pseudoglandular squamous cell carcinoma. Although it has been described in some processes unrelated to the use of chemotherapy (squamous cell carcinoma, pseudoepitheliomatous hyperplasia, keratoachanthoma, etc.), most studies associate it with chemotherapy, both systemic and due to extravasation or intradermal injection of the chemotherapy (2, 3). Although the pathogenesis of ESS remains unclear, it seems that it represents a non-specific reaction to damaged eccrine ductal epithelium induced by different stimuli. Chemotherapy-induced ESS could be explained by damage to the eccrine ducts, by excessive sweat gland production and/or drug concentration in this secretion. Moreover, other factors such as local irritation or friction may be implicated (1, 4). ESS does not require a specific treatment and it is not a prognosis factor; both depend on the underlying disease. It usually goes into remission spontaneously, but the use of topical or systemic corticosteroids may be useful if the lesions are itchy. The presence of ESS has rarely been described in patients receiving a haematopoietic stem-cell transplantation (after conditioning with chemotherapy) who go on to develop a graft-versus-host disease (GVHD), its coexistence with acute GVHD being exceptional (4). We present a case with acute GVHD coexisting with ESS.

CASE REPORT

The patient is a 50-year-old woman diagnosed with Hodgkin's lymphoma in 2010 who received chemotherapy for appoximately 6 months, with complete remission for a year and a half. In March 2012, she was diagnosed with secondary myelodyplastic syndrome that required an HLA-identical allogeneic transplant from a non-family donor after conditioning with busulfan and cyclophosphamide. As prophylactic treatment of the GVHD, the patient was given cyclosporine (2.5 mg/kg/12 h i.v. adjusted to maintain levels between 200–300 mg) and a brief course of methotrexate (15 mg day +1; 10 mg days +3, +6, +11). At day 7 post-transplantation, the patient started to suffer from mildly pruritic erythematous papules grouped in plaques on the back and chest, with a marked follicular distribution (Fig. 1). At day 11 post-transplantation, the patient complained of pain in the right hypochondrium, suggestive of hepatic GVHD, and was given treatment with intravenous methylprednisolone (40 mg/12 h i.v.). Both the cutaneous and hepatic symptoms improved notably with this treatment but the skin lesions worsened when the dose was reduced. At day 21 post-transplantation, a skin biopsy was performed showing signs of GVHD affecting the follicles, marked epidermal dysmaturation and focal lesions of ESS (Fig. 2). At day 51 post-transplantation, given the persistence of similar lesions in the sub-mammary region, abdomen and inner thigh, another biopsy was conducted with similar findings. The lesions disappeared completely one week after administering topical glucocorticoids and with maintenance of cyclosporine at the same dose.

DISCUSSION

The clinical and histopathological findings and their course were compatible with an acute follicular GVHD and ESS. As stated above, there are few reported cases of coexistence of GVHD and ESS. With chronic GVHD, effects on the eccrine sweat glands have been described in the form of destruction and fibrosis, but rarely as ESS and glandular dilatation (5, 6). With

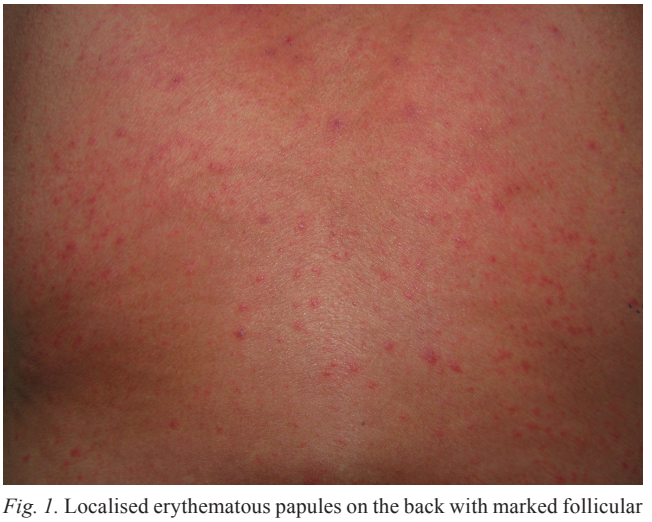

Fig. 1. Localised erythematous papules on the back with marked follicular distribution.

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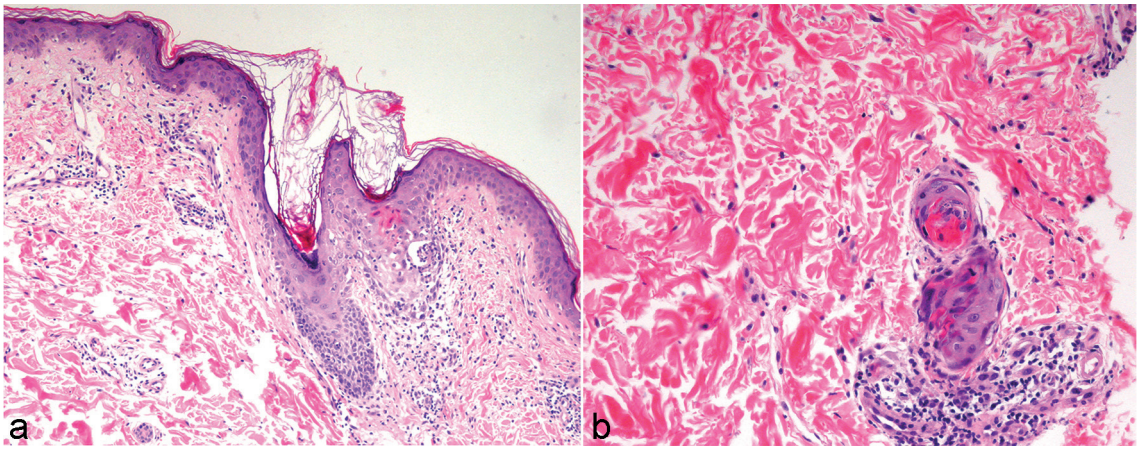

Fig. 2. Graft-versus-host disease affecting the follicles (a) and focal lesions of eccrine squamous syringometaplasia (b).

respect to acute GVHD, until 2003 no case of coexistence of both processes had been described, and it was even suggested that the appearance of ESS could be a diagnostic key to distinguish between a chemotherapy-induced medicinal reaction and acute GVHD (7). As well as the rarity of finding symptoms of ESS in a patient with acute GVHD, our case has other unusual characteristics, such as the early appearance of lesions and marked follicular distribution, which could correspond to an infrequent form described as follicular variant in acute and chronic GVHD (8, 9).

The authors declare no conflict of interest.

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