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CLINICAL REPORT

Superficial Mucoceles and Lichenoid Graft Versus Host Disease: Report of Three Cases

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Superficial mucoceles are subepithelial extravasations of sialomucin that occur at the epithelial-connective tissue interface and are directly related to minor salivary glands. They have been described in association with oral lichen planus and, exceptionally, with chronic graft versus host disease. Three patients who underwent an allogeneic bone marrow transplantation for a chronic myelogenous leukaemia presented multiple superficial mucoceles and an oral lichenoid graft versus host disease. ***Key words: allogeneic bone marrow transplantation; chronic myelogenous leukaemia.***

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Mucoceles are cysts of the minor salivary glands. They may present superficially and are then considered an independent variant. Superficial mucoceles are subepithelial extravasations of sialomucin that occur at the epithelial-connective tissue interface and are directly related to minor salivary glands (1, 2). They appear as asymptomatic vesicular lesions, single or multiple, and are most frequent in the soft palate, the retromolar region and the buccal mucosa (1, 4). They have been described in association with oral lichen planus (1, 4), and, exceptionally, with chronic graft versus host disease (GVHD) (5, 6). We describe three patients with multiple superficial mucoceles who also presented an oral lichenoid GVHD.

CASE REPORTS

Details of the three patients reported are given in Table I. All three patients had undergone an allogeneic bone marrow transplantation (BMT) (twice from the same donor in patient 2) for a chronic myelogenous leukaemia (Philadelphia$^+$) and had developed manifestations of a GVHD before the lesions appeared. For this they received different treatments with cyclosporin A and steroids (all patients), tacrolimus (patient 2), mycophenolate mofetil (patient 3) and thalidomide (patient 1). Between 5 and 10 months after the BMT, they showed multiple vesicular lesions on the mucous membrane of the lips or the soft palate. The lesions were asymptomatic, clear, tense vesicles 0.1 – 0.4 cm in diameter on clinically uninflamed mucosa (Figs. 1 and 2). All three patients referred to enlargement of the vesicles at mealtimes. On physical examination, all patients also showed net-like white striae on the buccal mucosa (all patients), the lips (patient 1) and the palate (patient 2) (Fig. 2). In all cases, the mucoceles remitted without treatment within a few weeks, but in patient 1 the lesions recurred one month later. Patient 1 died some months later from a pulmonary GVHD and complicating infections. Patients 2 and 3 are still alive and have not shown recurrences of the superficial mucoceles.

In all three cases, the histological study of the lesions showed a vesicle formation at the epithelial-connective tissue interface with partial epithelial regeneration across the vesicle floor. The vesicles were filled in by a mucoid material (PAS- and alcian blue-positive) and, surrounding the lesions, there was a mild lymphocytic infiltrate in cases 1 and 3, and in case 2, an intense lichenoid infiltrate, which also affected the gland ducts located under the vesicle (Fig. 3). In case 1, a minor salivary gland duct with cytologic atypia was present in close proximity to the vesicle (Fig. 4), but no cytologic atypia was found in patients 2 or 3.

In all cases, biopsies from the buccal mucosa distant to the mucoceles showed a lichenoid infiltrate (more severe in case 2) with hydropic degeneration of the basal layer and some necrotic keratinocytes suggesting an oral lichenoid GVHD.

DISCUSSION

Superficial mucoceles were first described by Eveson in 1988 (1), but since then they have rarely been reported in the literature (1 – 4). They are asymptomatic vesicular lesions usually located in the soft palate, in the retromolar pads, in the buccal mucosa and lips. They usually appear as multiple vesicular lesions and are unpredictably recurrent at irregular intervals, sometimes over many years (1, 2). In some cases the lesions seem to appear at mealtimes. Microscopically, they are subepithelial extravasations of sialomucin that occur

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Table I. *Details of the three patients with superficial mucoceles*

	Patient 1	Patient 2	Patient 3
Age (yr)/sex	38/male	53/male	21/male
Donor	HLA-identical sibling	HLA-identical sibling	HLA-identical non-related donor
Conditioning treatment	Busulfan+ cyclophosphamide	Busulfan+ cyclophosphamide	Cyclophosphamide+ TBI
GVHD (day post BMT)	Oral, hepatic, pulmonar (+ 95)	Hepatic, mucocutaneous, intestinal (+ 40)	Mucocutaneous (+ 75)
Superficial mucoceles			
Onset after BMT	5 months	10 months	5 months
Location	Lips	Soft palate	Soft palate
Associated disease	Lichenoid GVHD in the buccal mucosa	Lichenoid GVHD in the buccal mucosa and palate	Lichenoid GVHD in the buccal mucosa

BMT= bone marrow transplantation, GVHD= graft versus host disease, TBI= total body irradiation.

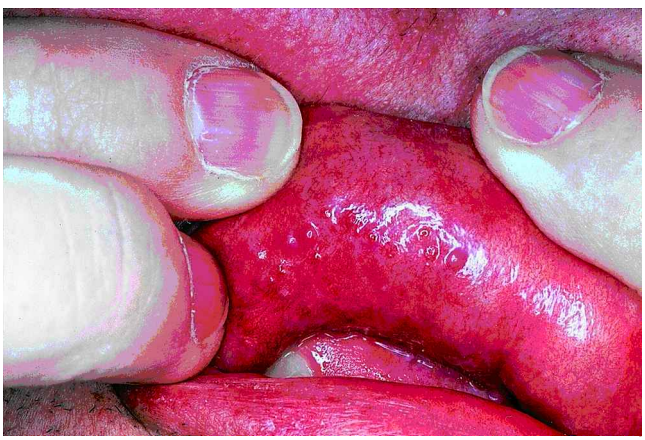

Fig. 1. Case 1. Multiple small vesicles on the upper lip.

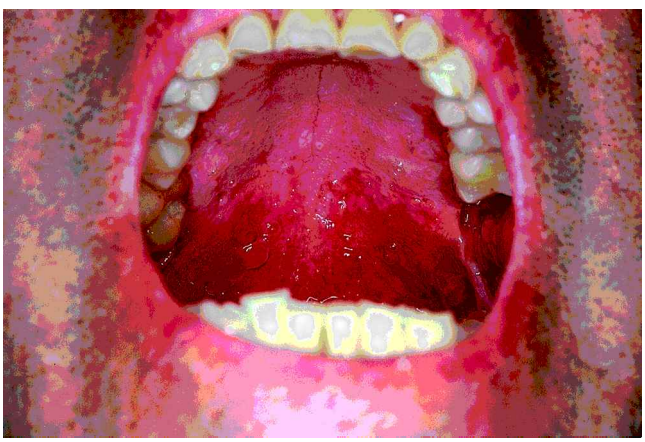

Fig. 2. Case 2. Multiple tense, bluish-translucent vesicles and net-like white striae on the palate.

at the epithelium-connective tissue interface. The epithelium of the roof is usually attenuated and, in most cases, there is partial or complete basal epithelial

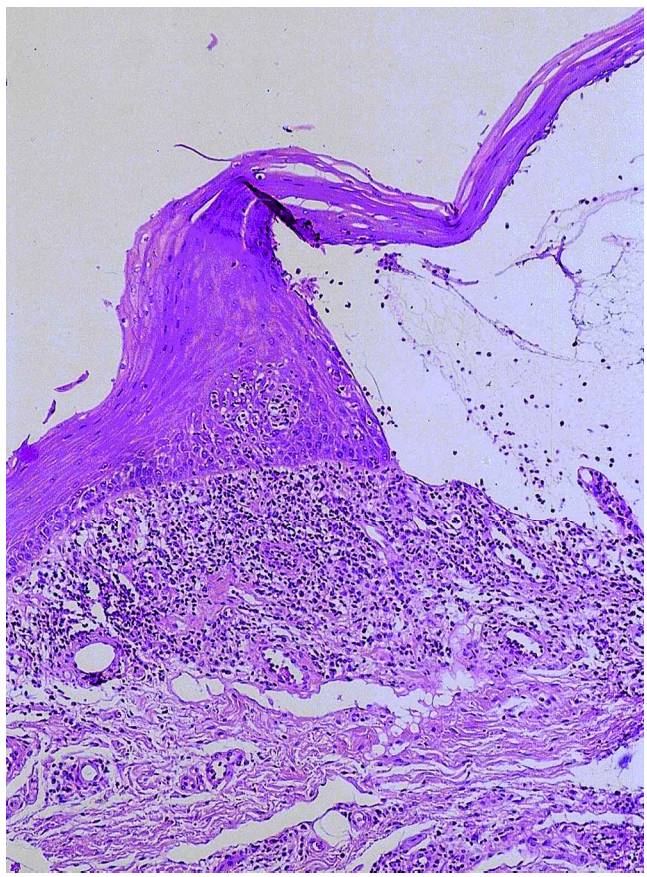

Fig. 3. Case 2. Superficial mucocele: a vesicle formation at the epithelial-connective tissue interface and a lichenoid lymphocytic infiltrate surrounding the lesion. Haematoxylin-eosin.

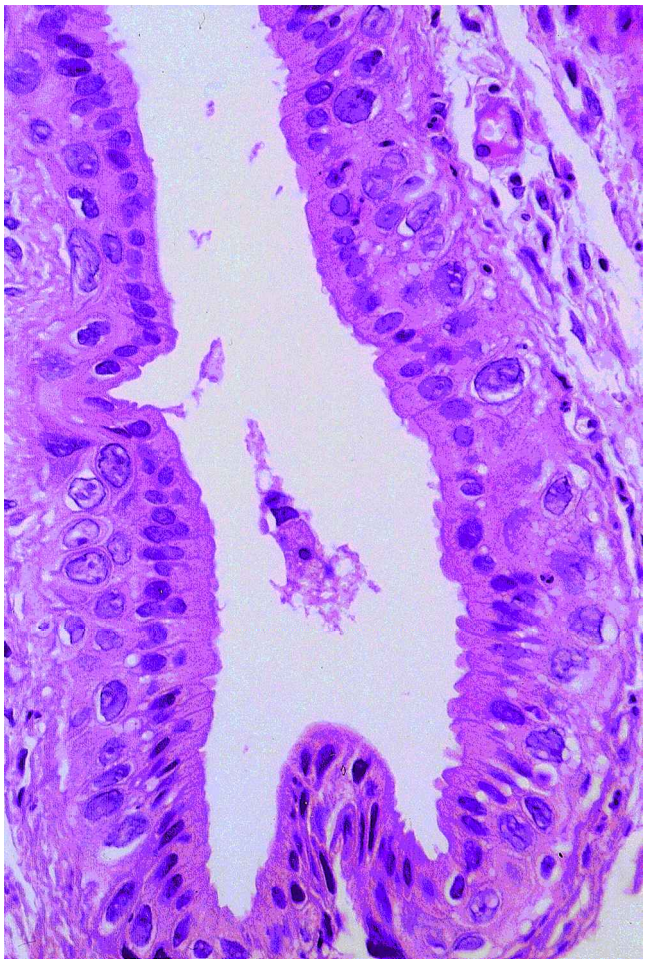

Fig. 4. Case 1. A minor salivary gland duct with cytologic atypia was present in close proximity to the vesicles. Haematoxylin-eosin.

regeneration. There are salivary gland ducts close to these vesicles. It is remarkable that, in the cases reported in the literature, five out of eight patients reported by Eveson (1) had a concomitant oral lichen planus, although in none of the cases was there evidence of mucosal disease in the biopsy specimens of the superficial mucoceles. Two out of four patients reported by Bermejo et al. (4) had concomitant lichen planus, and they suggest an aetiopathological mechanism similar to that involved in the miliaria crystalina. Our three patients with superficial mucoceles had undergone an allogeneic BMT and had clinical and histological evidence of an oral lichenoid GVHD. This association has been described in adults and paediatric patients (5, 6). In all of our patients, a lichenoid infiltrate was present in the buccal mucosa, clinically seen as white reticulated striae. In patient 2, the lichenoid infiltrate could be recognized surrounding and under a superficial mucocele. Although the aetiology of superficial mucoceles remains unknown, the association to a lichenoid infiltrate in the oral mucosa is probably not casual. Superficial mucoceles develop in the areas less exposed to traumas, and therefore the proposed aetiopathologic mechanism in conventional mucoceles of a trauma seems unlikely (2). A possible mechanism could be a duct obstruction or rupture caused by increased intraductal pressure in the intraepithelial portion of the duct (by mucus plugs, altered epithelial turnover or both) (2). In patients with chronic oral lichen planus or with oral lichenoid GVHD, the lymphocytic infiltrate could, at one stage, affect small salivary gland ducts, causing blockade or rupture of the duct and subepithelial collections of mucus (4).

Another point to remark is the cytologic atypia in the duct gland that we found in the first case. We did not observe atypia in the other two cases and we have not found any reports of superficial mucoceles with cytologic atypia in the literature. We consider that it could be secondary to a chemotherapy effect, as it has been described in other organs (7), and that it is probably unrelated to the mucoceles.

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