CASE REPORT

Oral Sweet's syndrome occurring in ulcerative colitis

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Accepted 3 March 2017

SUMMARY

A man aged 78 years presented with a 3-week history of tender mouth ulceration associated with arthralgia and weight loss. He had ulcerative colitis that was diagnosed 10 years previously which was well controlled on adalimumab 40 mg fortnightly. Biochemical and haematological investigations showed raised inflammatory markers (CRP 105) and a marked neutrophilia (10). On examination, the patient had severe oral ulceration involving the anterior tongue and lips. In addition, on cutaneous examination had tender erythematous nodules involving the forehead. Histology from a diagnostic punch biopsy showed marked dermal oedema with an inflammatory infiltrate consisting of neutrophils. Our working diagnosis was therefore oral Sweet's syndrome. The patient was then started on oral prednisolone and later received colchicine which led to a complete resolution of symptoms.

disease (IBD) first described by Robert Sweet in 1964.¹ It is characterised by the acute onset of fever, neutrophilia and tender erythematous skin lesions (papules, nodules and plaques) predominantly affecting the upper extremities, face and neck.² In the setting of IBD, Sweet's syndrome is more commonly associated with Crohn's disease than ulcerative colitis (UC), and our patient presented in the setting of UC. The onset of symptoms from Sweet's syndrome usually occur after the initial diagnosis of IBD, but may antedate the onset of IBD (21%) and has also been reported to occur 3 months after proctocolectomy in patients with UC.³ Early recognition and biopsy is important as it is very responsive to topical and oral corticosteroids. In addition, colchicine has been used frequently to obviate the need for steroids in the setting of corticosteroid dependence.

BACKGROUND

This case is interesting as our patient had oral Sweet's syndrome in addition to cutaneous involvement (figures 1–3). Sweet's syndrome is a rare extraintestinal manifestation of inflammatory bowel

CASE PRESENTATION

This man presented with an interesting collection of symptoms, including a 3-week history of tender mouth ulceration associated with arthralgia and weight loss. He had a history of IBD (UC), which was well controlled on adalimumab. He was

Figure 1 Ulceration involving the anterior surface of the tongue.

Figure 2 Ulceration involving the lips and tongue.

Figure 3 Complete resolution following treatment with colchicine.

To cite: Nestor LA, Tobin A-M. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2016-218249

otherwise well and had no previous dermatological or haematological conditions. On cutaneous examination, however, it was noted he had evidence of lesions that were consistent with Sweet's syndrome, later confirmed histologically. The diagnosis of Sweet's syndrome was made and our patient was treated successfully with corticosteroids and colchicine (figure 3).

DIFFERENTIAL DIAGNOSIS

Our other differential diagnosis was oral candidiasis (however, the patient was treated appropriately with antifungal therapy and this did not lead to resolution).

OUTCOME AND FOLLOW-UP

Initially, the patient was treated with oral corticosteroids and subsequently placed on colchicine which lead to complete resolution in symptoms.

DISCUSSION

Sweet's syndrome presenting in IBD is a rarity in itself; however, we report a case of oral Sweet's syndrome. It is important to note however in this patient that the biopsy was taken from a cutaneous lesion on the cheek. In addition to this, it is important to note that oral candidiasis was a likely explanation for the appearance of the oral mucosa. However, given that this did not respond to appropriate treatment of same, and given the rapid response to oral corticosteroids and colchicine, we have deduced that this is indeed an oral manifestation of Sweet's syndrome. Oral Sweet's syndrome has previously been reported in the literature⁴⁻⁶ and responded very well to oral corticosteroids.⁵ Indeed in one of these cases, the patient presented in a similar manner to ours with oral pustulosis and it has therefore been recommended that in the setting of recurrent oral ulcers with cutaneous lesions, Behçet's disease and neutrophilic dermatosis must be suspected, particularly as in our patients' case, where there is a history of IBD.⁶

Learning points

- ▶ Sweet's syndrome is an important differential diagnosis to consider in the setting of oral ulceration.
- ▶ Up to one-third of patients with inflammatory bowel disease (IBD) will experience extraintestinal manifestations involving the skin.
- ▶ Sweet's syndrome is a rare manifestation of IBD; however, if recognised early and treatment timely instituted, complete resolution can be achieved.

Contributors LAN, the principal author, is responsible for acquisition of data and analysis. A-MT reviewed and edited the article prior to submission.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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