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Case report

Oral ulcers as first clinical manifestation in Cronh's disease

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TO THE EDITOR

Oral mucosa has several functional similarities with small intestinal epithelium, including immunological functions. Oral features of Crohn's disease include ulcerations, lip fissuring, cobblestone plaques, recurrent buccal space abscesses and mucosal tags.[1,2] Recurrent apthous ulcerations and Crohn's disease share features such as familial tendency, psychological factors and response to corticosteroids and immunosuppresive drugs; both are considered to be of autoimmune nature.[3,4] Differential diagnosis of oral ulcers in Crohn's disease must be done mainly from Adamantiadis-Behcet disease and herpetic stomatitis and can sometimes be extremely difficult.[5]

A 21 year-old student presented at the Emergency Department of our hospital complaining of unbearable perianal pain during the previous 3-days. Physical examination showed an erythematous and perianal area extremely sensitive to touch. Digital examination was so painful that the patient could hardly stand it. However, no signs of hemorrhoid thrombosis or any kind of fissure were evident. Discussing past medical history including sexual medical history, the patient mentioned that 4 years earlier he had been diagnosed at another hospital with "atypical" Adamantiadis-Behcet disease, following the appearance of recurrent oral ulcerations. Opthalmologic examination was within normal limits. During those years no medical treatment had been started on as the patient did not fulfil the criteria of the typical Adamandiadis-Behcet disease. As the probability of a co-existing intestinal disease had to be excluded, despite the fact that the patient was symptomless, all routine examination tests were performed including colonoscopy. Terminal ileum was not examined during endoscopy. All biopsies from the large bowel failed to prove any specific or suggestive finding of inflammatory bowel disease. The patient was started on therapy with sulfasalazine suppositories and cefuroxime axetil per os resulting in complete symptom remission within a 3 days time. A second colonoscopy at this hospital with terminal ileum biopsies was performed, which finally led to the diagnosis of Crohn's disease with terminal ileum and oral cavity involvement.

Key words: Crohn's disease, Adamantiadis-Behcet disease, oral ulcers, colon ulcers, metastatic Crohn's disease

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Cronh's disease with Adamandiadis-Behcet's disease-like appearance has been rarely reported[6]. In addition, many Adamandiadis-Behcet's disease patients have been reported with evidence of gastrointestinal involvement which was identical or extremely similar to that of inflammatory bowel disease patients.[7,8] In addition, one case with these two diseases co-existent has also been reported.[9]

Gastrointestinal involvement in Adamandiadis-Behcet's disease is characterized by ulceration along the gastrointestinal tract including the esophagus and most frequently occurs in the terminal ileum and cecum.[10-12] Although enteroclysis was highly suggestive of terminal ileum involvement in the patient, no further investigation had been performed. Thus, for four years, the patient was followed up but no treatment recomended, as the diagnosis remained "atypical".

In the patient presented here, oral ulcer biopsies were not suggestive of Cronh's disease. However, granulomas in biopsy specimens should not always be considered as

the hallmark of Crohn's disease according to our own experience from bowel biopsy studies. This presumably may also happen to biopsies from extraintestinal tissues with high suspicion of the so-called "metastatic" Crohn's disease to sites other than the bowel. When the gastrointestinal tract is involved in a patient with Adamandiadis-Bechet's, the differential diagnosis from Crohn's disease is only of aetiologic importance, as the pharmacologic treatment used is similar.[13-15] Oral Cronh's disease can be misdiagnosed as Adamandiadis-Behcet disease for many years, until correct diagnosis is made. Oral ulcers, along with other extraintestinal manifestations, especially these of skin, may be active despite remission of bowel symptoms and often require additional local therapy.

Thus, in any patient with recurrent atypical oral ulcerations or recurrent oral cavity apthae of unknown aetiology, the possibility of Crohn's disease should never be ignored or even underestimated, especially in younger patients. Furthermore, in any such case of atypical Adamandiadis-Behcet disease, the gastrointestinal tract must always be carefully investigated in order to exlude other diseases, including Adamandiadis-Behcet disease, with gastrointestinal involvement, or even quiescent Cronh's disease.

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