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A Case Report of Recurrent Aphthous Stomatitis: A Difficult Case

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ABSTRACT - Recurrent aphthous stomatitis (RAS) is one of the commonest disorders of the oral mucosa, with the onset of recurring painful ulcer lesions typically occurring in childhood or adolescence. The disorder is divided into three clinical forms: ulcer minor, ulcer major and herpetiform ulcer, the first form being the commonest. The etiology remains undefined, but there are evidences of several predisposing factors and pathogenesis points to a disorder of an immunological nature. Diagnosis is based on the clinical history and physical exam of the patient and the histopathological exam yields unspecific findings. Case Description: The authors report a case of RAS that was difficult to control, in which the coexistence of predisposing factors such as family history and psychosomatic factors appeared to make it more difficult to control the disease. Discussion: Several drugs are mentioned for treating RAS, and the most effective of these have adverse effects that restrict their use. Although the etiology remains undefined, the coexistence of predisposing factors, such as family history, contribute to more severe conditions, with great impact on the quality of life. This emphasizes the importance of lasting and multidisciplinary follow up, in order to ameliorate the great social and psychological impact, with more attention being paid to the predisposing factors in each case.

KEYWORDS - Recurrent aphthous stomatitis, mouth ulcers, aphthae

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Introduction

Recurrent aphthous stomatitis (RAS) is characterized by the presence of painful ulcer lesions in the oral mucosa, frequently multiple, round or oval, with an erythematous halo, usually with onset in childhood or adolescence (1) and decreasing incidence and severity of lesions with age (2).

Prevalence ranges from 2% to 66% of world population (3,4), being three times commoner in white Americans than in Afro-Americans (1,5). The etiology still remains undefined (1,6), but there are evidences of several predisposing factors, such as nutritional deficits (vitamin B12, folic acid and iron), hormonal alterations, stress, local trauma, food allergy, genetic factors (6), and bacterial and viral infections (7). Pathogenesis points to a disease of a basically immunological nature (2,3), mediated by auto-antibodies and alterations in cell immunity (1,6).

Recurrent aphthous stomatitis may present in three clinical forms: aphthous ulcer minor (Mikulicz's aphtha), aphthous ulcer major (Sutton's ulcer, periadenitis mucosa necrotica recurrens) and herpetiform aphtha. The aphthous ulcer minor is the commonest form, accounting for 80% of cases (3). It mainly affects the nonkeratinized oral mucosa, most frequently affecting the lip and jugal mucosa, followed by the ventral part of the tongue, floor of the mouth and soft palate (3). It is the form of clinical presentation of shortest duration (around 7 to 14 days) and there may be single or several, round or oval lesions measuring from 0.5 to 1cm, with a maximum of six lesions at a time. Major aphtha presents deeper lesions,

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larger than 1cm that are capable of leaving scars, and its duration ranges from 2 to 6 weeks. The number of lesions may range from 1 to 10. Whereas, the herpetiform ulcers are much rarer. It presents various small, painful ulcers, measuring from 1 to 3 cm and are called herpetiform because of their clinical similarity to herpetic stomatitis lesions. The lesions may fuse and produce much larger ulcers, with a duration of 7 to 10 days, on an average (3).

The diagnosis of RAS is based on the patient's history and physical exam (1,2), and the anatomic-pathological exam yields unspecific findings (3,5). Other systemic diseases that may manifest as ulcerations in the oral mucosa must be ruled out, such as the PFAPA syndrome, HIV (1,8), Behcet's syndrome (1,8,9), Crohn's disease, celiac disease, gluten-sensitive enteropathy, Reiter's syndrome and cyclic neutropenia (1,2,4).

The aim of treatment is to resolve the lesions, control pain and recurrence of ulcers (6). This includes oral hygiene and dietary measures, as well as a varied therapeutic arsenal, as well as a varied therapeutic arsenal, selected according to the severity of the case, and includes topical and systemic medications. Topical corticosteroid continues to be the most used therapy in several institutions (10,11,12). Chlorhexidine is also mentioned for topical use to improve bacterial control and it may reduce the duration of the ulcer and increase the number of lesion-free days (1). Systemic corticosteroids are used during the acute stages of more severe cases with doses ranging from 20-60mg/day. Other systemic medications, such as pentixifylline, dapsone, colchincine, levamisol and thalidomide are used. Thalidomide has been shown to be the most efficient drug for treating RAS (2,5,13), with remission of lesions in around 50% of patients. However, the severity of its adverse side effects such as sedation, neutropenia, neuropathy and teratogenicity have limited its indication.

Case Report

The patient L.O.D, a fifteen-year-old white girl, student, resident in the Federal District in Brazil, consulted the Stomatology Clinic of the Dentistry Course at the UnB, on August 7th, 2008, accompanied by her mother, complaining of recurrent aphthae in the oral cavity over the last four years. She reported having sought medical attendance during the worst periods of the condition, and had used several unspecified medications. At the time of the first consultation, she had made use of 20mg omeprazol, prescribed for 30 days due to the diagnosis of edematous reflux esophagitis and moderate enanthematous pangastritis, proved by upper digestive endoscopy on July 17th, 2008. She denied having any other symptomatology or previous diseases. In the family history, she reported that her father also had recurrent aphthae.

The patient was assessed by the clinic dentist and otorhinolaryngologist. On oral clinical exam, she presented four minor aphthae lesions distributed throughout the jugal and lip mucosa and left lateral edges of the tongue, and a major aphtha in the jugal mucosa on the right and another on the left, with areas of fibrosis close to the ulcer regions (Figures 1-4). There was presence of abundant bacterial plaque, caries in some teeth and root remainders corresponding to teeth 14 and 17.

Prednisolone (40mg) for seven days, dexamethasone elixir (0.5mg/ml) for

Figure 1. Major aphthous ulcer, jugal mucosa.

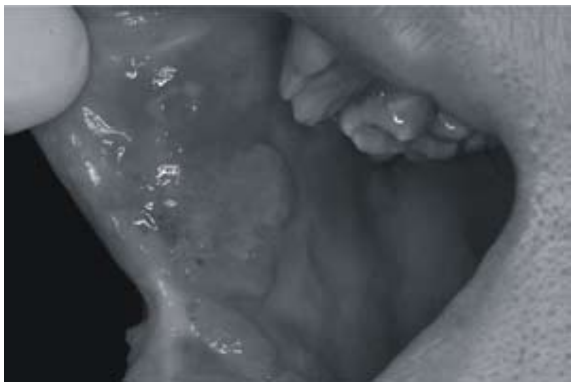

Figure 2. Major aphthous ulcer of longer than 6 weeks' duration, jugal mucosa.

Figure 3. Minor aphthous ulcers, tongue.

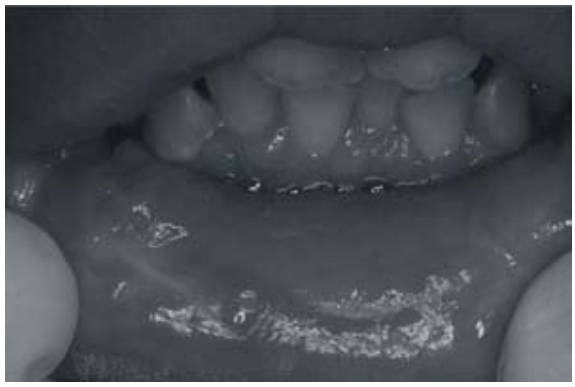

Figure 4. Minor aphthous ulcers, lower labial mucosa.

mouthwashes three times a day for 10 days and 0.12% Chlorhexidine for mouthwashes twice a day for fifteen days were prescribed, in addition to providing guidance as regards diet and oral hygiene. When she returned for follow-up on August 14th, 2008, she showed significant improvement in the symptoms and clinical aspects of lesions. Prednisolone (5 mg) was prescribed for a further seven days. After performing prophylaxis and sub- and supragingival scraping, the patient was referred to the dentist for caries treatment and extraction of the indicated teeth. The patient was instructed to return to the service if the condition recurred.

On October 16th, 2008, the patient returned with recurrence of the symptoms, presenting two major aphthae on the jugal mucosa and three minor aphthae in the lip mucosa and floor of the mouth. New therapy was prescribed, including topical and systemic corticosteroid (Dexamethasone Elixir and 40 mg Prednisolone for five days) 5 mg/day folic acid for an indefinite time, low level laser therapy on the lesions and biopsy of some of the lesions. Laboratory exams were requested (complete hemogram, glucose, seric levels of iron, Gama GT, amylase, alkaline phosphatase, glucose, urea, creatinine, GTO/AST, TGP/ALT and iron). Biopsy showed unspecific inflammatory infiltrate. Laboratory exam results were normal. The new upper digestive endoscopy performed by pediatric gastroenterologist showed persistence of pangastritis and discarded other gastrointestinal inflammatory diseases.

The patient began to receive weekly control (during improvement periods every 15 days) at the Stomatology service at HUB, and 10mg prednisolone was prescribed as a maintenance dose. However, persistence of minor aphthous lesions was observed, without healing of the two major aphthae. In the view of the patient's introspective, sad look and unstable condition of the family environment due to her father's alcoholism, according to the mother's report, psychotherapy treatment was instituted at the Psychology Service of HUB.

In the view of this developmental condition, the systemic corticosteroid was withdrawn on April 4th, 2009, and the use of Pantoprazol 80mg/day for 30 days and complex B, 1 capsule a day, for 30 days were prescribed; laser therapy, folic acid and psychotherapy were maintained. On April 4th and 16th, 2009, these measures showed significant improvement of the condition, diminishment of pain and better acceptance of diet. The clinical exam showed healing halo edema around the lesions In spite of the improvement observed, weekly consultations continued in order to confirm control of the condition, or the need to reassess the treatment procedure.

Discussion

RAS is a disease with great impact in the quality of life (4,14,15) because the discomfort and pain caused by the lesions make it difficult to control diet and perform oral hygiene and favor the appearance of caries. Furthermore, studies have shown the impact on the social and psychological life of patients affected by the disease (15). For these reasons, patients need a multidisciplinary follow up in order to obtain careful investigation and better control of the condition. Because the etiology of this disease is still unknown, therapy is not specific and its

efficiency is limited (1). More attention must be given to the predisposing factors.

In the reported case, positive family history appears to contribute to the severity of the lesions and difficulty in controlling them. Studies have shown that there is greater and more severe prevalence in patients whose family members are also affected, when compared with the general population with RAS (1). The patient's emotional state also seems to contribute to the severity of the condition, since some studies have indicated that that the emotional state interferes in the therapeutic response (15).

Complex B and folic acid were prescribed although some studies have shown infrequent resolution of the condition with the use of vitamin supplements. Whereas, other studies have shown that hematologic deficiencies of iron, folic acid and vitamin B12 were two times commoner in patients with RAS than in the control group (1,16). Pantoprazol was prescribed with the aim of gaining better control of pangastritis, and laser therapy was indicated to stimulate the mucosa healing (17, 18, 19). Systemic corticosteroid was used in the acute phases and when pain worsened, since studies have proved their efficacy in reducing the symptoms (1).

Final Considerations

Although the etiology of RAS is still unknown, the interaction among immunological, genetic and psychosomatic factors is strongly involved in the appearance and severity of the lesions. The clinical case reported unites these factors, and although the therapy applied provided significant improvement in the condition, long term follow up of the patient is necessary to ensure adequate control of the disease, and further studies are required to confirm the efficiency of the treatment.

Resumo

GUIMARÃES, Maria Cristiane Machado, GUIMARÃES, Maria do Carmo Machado, LEITE, André Ferreira, FIGUEIREDO, Paulo Tadeu de Souza, MELO, Nilce Santos de. Relato de Caso de Estomatite Aftosa Recorrente: Um Caso Difícil. Oral Sci.,Jan/Apr. 2010, vol.2, no.1, p. 43-47.

A estomatite aftosa recorrente (EAR) é uma das doenças mais comuns da mucosa bucal que cursa com lesões ulcerosas dolorosas que se iniciam geralmente na infância ou adolescência. A doença apresenta três formas de apresentação clínica: afta menor, afta maior e afta herpertiforme, sendo a primeira a forma de apresentação mais comum. A etiologia permanece indefinida, mas há evidências de diversos fatores predisponentes e a patogenia aponta para uma doença de caráter imunológico. O diagnóstico baseia-se na história clínica e no exame físico e o histopatológico é inespecífico. Apresentação do caso: Os autores relatam um caso de EAR de difícil controle em que a coexistência de fatores predisponentes como antecedentes familiares e psicossomáticos, parecem dificultar o controle da doença. Discussão: Diversas drogas são citadas no tratamento da EAR. As mais efetivas apresentam efeitos adversos que restringem o uso. Apesar da etiologia permanecer indefinida, a coexistência de fatores predisponentes, como história familial, contribuem para quadros mais severos, com grande impacto na qualidade de vida. Isso realça a importância de um acompanhamento duradouro e multidisciplinar, a fim de amenizar o grande impacto social e psicológico, dando maior atenção aos fatores predisponentes em cada caso.

Palavras-chave - Estomatite aftosa recorrente, úlcera bucal, afta.

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