

Orofacial Crohn's Disease: A Case Report

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ABSTRACT

Crohn's disease is a chronic disease of the digestive system. It is characterized by lesions predominantly located in the small intestine and colon, although they may also occur in any segment of the gut, including oral cavity. Involvement of the oral mucosa in Crohn's disease may be underreported, as up to 42% of the pediatric patients with Crohn's disease were found to have oral lesions after undergoing a thorough oral examination. Here, we present a case of Crohn's disease, in which the patient referred to the dentist due to non-healing aphthous ulcers in the mouth. Our patient, a 16 year-old boy, admitted to the dentistry clinic with swelling of the oral mucosa and the lips, for the last 3 months. The patient was referred to our department due to non-response of the mucosal lesions to repeated cycles of medical treatment. Colonoscopy revealed a cobblestone appearance especially in the left colon, partly normal mucosa, and exudative ulcers. Biopsy samples showed increased inflammatory cell infiltration in the lamina propria, cryptitis in some of the crypts. A close collaboration between gastroenterologists and dentists is useful when addressing the diagnosis and appropriate management of these patients.

Keywords: Crohn's Disease, Oral Cavity, Dentist

Introduction

Crohn's disease is a chronic disease of the digestive system. It is characterized by lesions predominantly located in the small intestine and colon, although they may also occur in any segment of the gut, including oral cavity (1). Due to the prolonged course of the disease, diagnosis may be problematic; however the findings of lesions in the oral mucosa may help to raise suspicion. The clinical spectrum of orofacial CD includes hyperplasia, cobble stoning, ulceration of the buccal and gingival mucosa and swelling of the lips and face. Involvement of the oral mucosa in Crohn's disease may be underreported, as up to 42% of the pediatric patients with Crohn's disease were found to have oral lesions after undergoing a thorough oral examination (2). Here, we present a case of Crohn's disease, in which the patient referred to the dentist due to non-healing aphthous ulcers in the mouth.

Case Report

Our patient, a 16 year-old boy, admitted to the dentistry clinic with swelling of the oral mucosa and the lips, for the last 3 months. The patient was referred to our department due to non-response of the mucosal lesions to repeated cycles of medical treatment. We were informed of the patient's history of anal fissure and diarrhea complaints 1 year ago. There were no features in the medical history of the patient and his parents, and the patient has 2 healthy siblings. In physical examination, weight was 55 kg (25-50 p), height 174 cm (50-75 p), cardiac pulse 96/min., and TA 120/80 mm/hg. We observed a cobblestone appearance inside the mouth and swelling in the lips. His anal examination revealed two fissures. In the examinations for definitive diagnosis of inflammatory bowel diseases (IBD), sedimentation was found 60 mm/h, and CRP 3.5mg/L. The patient's liver and kidney function tests were normal.

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Received: 21.09.2018 Accepted: 07.01.2019

Immunoglobulin G, A and M levels were 1260 mg/dl, 390 mg/dl and 66.9 mg/dl, respectively. Whole blood count analyses were Hb: 10.4 gr/dl, Htc: %32.2, PLT: 304000, Fe: 40 mg/dl and ferritin: 49 ng/ml. Upper and lower gastrointestinal system (GIS) endoscopies were performed on the patient with suspected inflammatory bowel disease. Colonoscopy revealed a cobblestone appearance especially in the left colon, partly normal mucosa, and exudative ulcers. Biopsy samples showed increased inflammatory cell infiltration in the lamina propria, cryptitis in some of the crypts (Figure 1). Buccal biopsy offer was not consented by the teenage boy. Directed by the patient's medical history, physical examination, laboratory and biopsy findings, the patient was diagnosed as Crohn's disease. The disease was extensive with PCDAI score of 30 and methylprednisolone treatment (60 mg/day) and mesalazine (40mg/kg/d) were started. During the observation period of two weeks, acute phase reactants of the patient normalized completely, cobblestone appearance was restored apparently and swelling recovered. The dosage of methylprednisolone was decreased by 5 mg per week for four weeks, and 2 mg/kg/day of azathioprine was added to the treatment. The patient has been followed with azathioprine maintenance in clinical and laboratory remission for the last 18 months.

Discussion

We report a case of a patient with Crohn's disease presenting with cobblestone-like oral lesions. We confirmed the diagnosis of Crohn's disease by performing colonoscopy. Oral lesions immediately responded to high dose steroid treatment.

A wide variety of disease-specific oral lesions have been described in patients with intestinal Crohn's disease. These

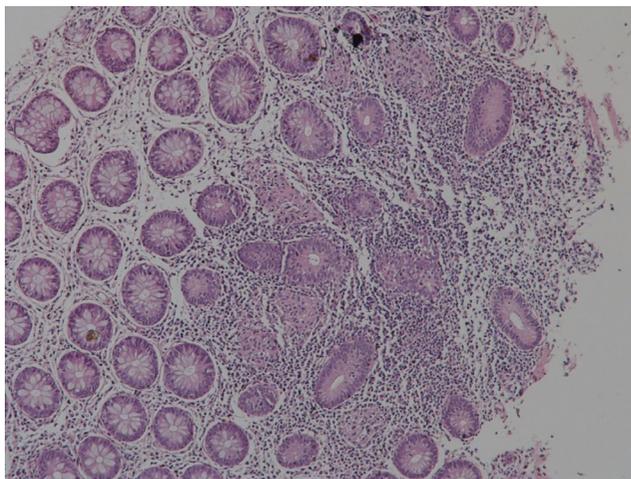


Figure 1. Histopathological examination (colon). Explanation: Hematoxylin eosin staining x10 magnification. In lamina propria; non-caseating microgranulomas, increased infiltration of focal lymphocytes and cryptitis (focal active colitis)

include swelling of the lips and buccal mucosa, cobblestoning, mucogingivitis, deep linear ulceration or mucosal tags (3).

The prevalence of oral manifestations in patients with Crohn's disease varies between 0% and 9% (4) in adults; however, it is more prevalent in children. In a prospective study of 48 children presenting with Crohn's disease, clinicians accurately identified the presence of oral CD in less than 50% of children with disease-specific lesions.

Studies with larger sample sizes had also suggested that the finding of certain oral lesions may be indicative of Crohn's disease, especially in children. In a prospective study, in a study conducted in Brazil comparing 62 Crohn's disease patients with a control group, oral lesions were found in 14.5% of the patients with Crohn's disease and in 9.6% of the control group, showing a low prevalence and an insignificant difference between the patients and controls (5). Colella et al, assessing the oral mucosa of 100 adult patients with Crohn's disease and ulcerative colitis for a period of 5 years, observed orofacial findings in only 7 patients, concluding that the association between Crohn's disease and orofacial findings is not as strong as it is reported (6).

Some oral lesions have been postulated as possible indicators of the existence of Crohn's disease. Rehberger et al. (4) described the case of a 20 year-old patient with painful intra-oral lesions. On endoscopy extensive lesions of the gastrointestinal tract were seen, and biopsies confirmed the diagnosis of Crohn's disease. In our case, we suspected Crohn's disease due to non-healing painful oral lesions and confirmed the diagnosis by colonoscopic biopsies.

There is no evidence-based algorithm for the treatment of orofacial Crohn's disease. Elemental diets appear to have variable outcomes. One case of facial and ileocolic CD showed improvement of disease with total nutrition (7). Several case reports document remission of oral symptoms with topical or systemic steroids used in conjunction with ASA or MP (8, 9). There have also been several cases of orofacial CD refractory to steroids which were treated with infliximab. (10). Since our patient had extensive disease systemic corticosteroids were applied as first line treatment according to the ECCO guidelines (11).

Many patients, particularly children, have involvement of the mouth when presenting with CD. Although usually subclinical, self-resolving and not requiring specific treatment, these disease-specific manifestations commonly harbor diagnostic material. Expert evaluation of the oral cavity is a useful adjunct in patients presenting with suspected IBD. A close collaboration between gastroenterologists and dentists is useful when addressing the diagnosis and appropriate management of these patients.

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