CASE STUDY

From Diagnosis to Management of Oral Manifestations Associated with Crohn's Disease: A Case Report

Mrunal Meshram[1], Rahul R. Bhowate[1], Vrushali Zamre[1], Vidya Lohe[1], Unnati Shirbhate[1], Kumar Gaurav Chhabra[2]

[1]Department of Oral Medicine and Radiology, Sharad Pawar Dental College and Hospital, Datta Meghe Institute of Higher Education and Research, Wardha, Maharashtra, India, [2]Department of Public Health Dentistry, Nims Dental College and Hospital, Nims University, Jaipur, Rajasthan, India

Abstract

In addition to the intestinal manifestations, the inflammatory bowel disease of Crohn's disease (CD) is also associated with some extra-intestinal symptoms, such as ulcers in the oral cavity, which tend to aggravate the general care and compromise the patient's general well-being. The following case presents a 23-year-old patient with a CD complicated by severe oral symptoms persisting despite early treatments, including recalcitrant recurrent aphthous stomatitis, mucosal swelling, and gingivitis. The patient was treated using a multidisciplinary approach that integrated both gastroenterology and dental aspects. Systemic corticosteroids, immunosuppressive therapy, topical corticosteroids, antibacterial mouth rinses, dietary advice, and general support were among the treatment modalities provided. This comprehensive plan, which significantly enhanced the patient's quality of life, also demonstrated improvement in oral health and a decrease in signs and symptoms. This case demonstrates how interdisciplinary strategies can help in CD oral manifestations management, given the enhancement of the patient's health. Further investigation and case studies are needed to refine treatment strategies and expand understanding of these oral lesions and concerns regarding them.

Key words: Crohn's disease, Oral ulceration, Inflammatory bowel disease, Oral manifestations of gastroenterological disorders

INTRODUCTION

Crohn's disease (CD) and ulcerative colitis are the two forms of inflammatory bowel disease (IBD) that are probably due to innate inflammation in the gut. The main cause of the condition, as well as its origins, is not clear. It is perhaps associated with the erosion of the mucous membrane. An individual who has a genetic predisposition to developing an immune inflammatory response to flora may have an alteration of his immunity tolerance and intestinal epithelial barrier function.[1-3] Lesions due to CD may occur anywhere along the gastrointestinal tract from the mouth to the anus. The anal canal and oral cavity are all parts of the digestive tract that may be affected by CD. Such cases may result in broad lesions, for example, in operating and severe intestinal wall penetration hyperplasia where chronic deep ulcerative enteritis organizes without domestic fiber-matrix effects. There may be infiltration and necrosis between normal and these lesions distal disorders. Complications such as fistulas, abscesses, and strictures all arise due to the inflamed peptic tissue in CD, bringing in more challenges in management.[4,5]

The most common symptoms in the patients are periumbilical abdominal pain and distress and watery stools that are also associated with chronic wasting and fever, malaise, and loss of appetite.[1,4,5] There is a low level of oral involvement in cases of CD, as only 20% is present, and in some instances, it can occur in advance of enteral involvement.[4,6,7] Orofacial granulomatosis (OFG) is a term used when there is hypertrophy of the orofacial region, specifically the lips, due to a granulomatous inflammatory process. The OFG

Address for correspondence:
Dr. Mrunal Meshram, Department of Oral Medicine and Radiology, Sharad Pawar Dental College and Hospital, Datta Meghe Institute of Higher Education and Research, Sawangi (M), Wardha, Maharashtra, India.
E-mail: mrunal.oralmed@dmiher.edu.in

Received: 09-11-2024
Revised: 24-12-2024
Accepted: 31-12-2024

was observed in systemic conditions such as sarcoidosis and CD.[8,9] In the following case of OFG and one case of pyostomatitis vegetans diagnosed as manifestations of CD have been presented, which focuses on clinical and oral manifestations, diagnostic criteria, and common management approach toward a patient diagnosed with CD, the breadth of treatment options available for OFG and pyostomatitis vegetans, focusing on the contributions of oral health-care providers to the recognition and treatment of oral complications of systemic diseases.[10]

CASE PRESENTATION

A 23-year-old female reported to the Outpatient Department of Oral Medicine with the chief complaint of painful ulceration in her mouth for 6 months. First, it was reported that the patient had lengthy diarrhea, frequent stomach cramps, and broad abdominal discomfort around 6 months ago. In addition to these symptoms, there was significant weight loss, frequent episodes of fever, and menstrual irregularities. Occasional oral pain was experienced by the patient, which was aggravated later by intake plus the increasing intensity during eating. Afterward, buccal mucosal ulcers intensified, accompanied by gingival bleeding, resulting in more discomfort and pain. Vestibular and posterior ulcers were developed in her soft palate. A systemic illness with consequences for the mouth is indicated by her concomitant conditional complaints; they demonstrate systemic and local manifestations resulting from an interaction between them. The very far-advanced manifestation of many systemic symptoms – including gastrointestinal distress, irregular menstruation, and painless mouth ulcerations – precludes such pathology as later stages rather than those typical at onset (e.g., CD or ulcerative colitis) for most patients present firstly with incomplete forms before progressing into complete ones.

A clinical examination of the patient revealed marked oral cavity irritation and erythema, with associated small pustules ranging from white to yellow in color and prominent tissue folding with generalized mucosal edema. It also showed deep, linear ulcers having the appearance of "snail tracks" at many sites – i.e., involving left commissural areas, right commissural areas, the left lateral border of the tongue, the right retromolar region and right up to the posterior-most portion of the hard palate. These were accompanied by thicker inflammatory gingiva having a cobblestone appearance in texture along with such type of ulceration accentuating this texture which gave way to erythematous patches. Erythematous areas could be observed over the palate and gingivae. No skin lesions were noted on inspection, despite the marked oral signs [Figures 1 and 2].

The figure describes the existence of ulcers in a few distinct oral cavity locations, suggesting a potentially serious clinical condition. The right buccal mucosa, or inner lining

Figure 1: Ulceration seen in the oral cavity. (a) Right buccal mucosa, (b) Left buccal mucosa, (c) Labial mucosa, (d) Gingiva in 15 region

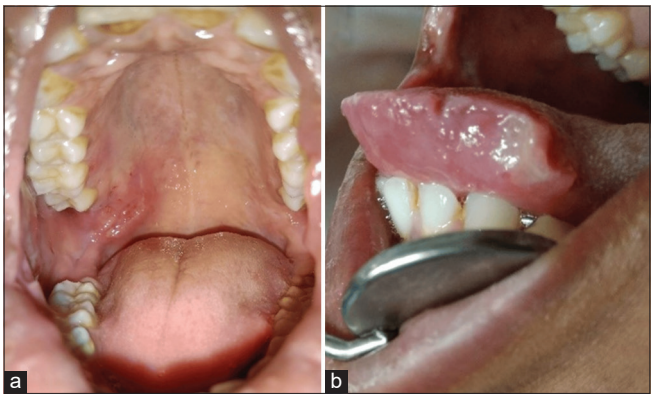

Figure 2: Ulceration seen on (a) palate and (b) tongue

of the cheek on the right side, is where ulcerations are seen; surrounding the ulcers, this area may show signs of necrosis, swelling, or redness. The symptoms on the other side of the mouth are mirrored in the damaged left buccal mucosa, indicating a bilateral problem or systemic ailment. Moreover, ulcers on the labial mucosa, or the inside surface of the lips, can cause discomfort and make it difficult to speak or eat. The gingiva in the 15 regions, which is often the upper left third molar, shows localized ulceration surrounding the gum tissue connected to tooth number 15.

Potentially resulting from infections, injuries, or inflammatory disorders, the palate ulcerations can be painful and interfere with eating and speaking. These ulcers, which are frequently the result of irritations or systemic problems, can cause discomfort and alter taste on the tongue. Both regions have ulcers, which raises the possibility of a more serious oral health issue that may require more research and care.

Pyostomatitis vegetans was the first diagnosis established because of oral lesions' characteristic "snail track" appearance. Pustular and ulcerative changes in oral mucosa are features of this disease. Pseudomonas vulgaris and

vegetans, which both have similar oral lesions and for whom a distinct consideration has to be made to give proper treatment (this underscores the importance of an accurate diagnosis), must also be included in the differential diagnosis. Hence, immunological investigations and histologic examination are needed to confirm it and exclude these other disorders. An incisional biopsy was taken from the affected area. The specimen was analyzed in ×10 after hematoxylin and eosin staining, which revealed underlying connective tissue and was infiltrated with mixed inflammatory cells – mainly lymphocytes as shown in Figure 3.

Several important aspects are revealed by the histological examination of oral cavity ulcers at ×10 magnification. The ulcerated area can be seen in the tissue architecture if there is a rupture in the epithelium. There may be signs of inflammation surrounding the ulcer, such as a mixed infiltration of neutrophils, lymphocytes, and plasma cells, which could indicate an immunological reaction to an injury or infection. In addition to probable necrotic debris inside the ulcer, the borders of the ulcer may exhibit hyperplasia of the epithelium during its healing process. In addition, you may see edema and increased vascularity in the connective tissue, which are signs of active inflammation. The overall histopathological results emphasize the mechanisms of tissue injury and repair linked to oral cavity ulcers. Therefore, the diagnosis of pyostomatitis vegetans was confirmed. As a result of the known association between pyostomatitis vegetans and chronic IBD, the patient was referred to a gastroenterologist and gynecologist. The investigation revealed pelvic inflammatory disease with cervix hypertrophy.

For the management of oral lesions, intralesional steroids (0.2% lignocaine and triamcinolone acetonide) are given in each visit weekly follow-up with systemic medication advised by the gastroenterologist and gynecologist. To treat flare-ups and decrease inflammation, a gynecologist and gastroenterologist may recommend systemic drugs, such as prednisone, for a patient with CD who presents with oral lesions. To keep inflammation under control and preserve remission, immunosuppressants such as mercaptopurine or azathioprine may be utilized. Targeting certain immune system pathways, biological medicines such as infliximab or adalimumab are frequently recommended for moderate-to-severe forms of CD. 5-aminosalicylic acid molecules, including mesalamine, have also been shown to have anti-inflammatory properties. Antibiotics such as ciprofloxacin or metronidazole may be used if a secondary infection is a concern. When a patient has trouble eating because of oral lesions, nutritional support – such as enteral nutrition or nutritional supplements – is crucial. To treat digestive issues and avoid consequences linked to acid reflux disease, proton pump inhibitors may be suggested. To treat linked disorders, the gynecologist may also prescribe hormonal therapy if necessary. Thus, in collaboration with the patient's health-care team, it is essential to customize the treatment plan based on the patient's unique demands and general health state. After the 3rd week, the lesion healed completely as shown in Figure 4.

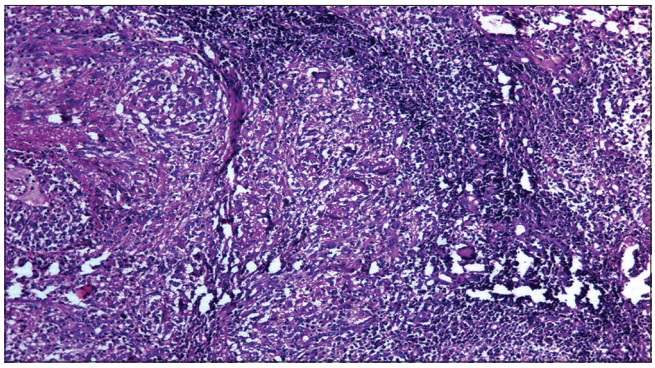

Figure 3: Histopathological view of oral cavity ulceration in ×10 magnification

DISCUSSION

The CD is a chronic inflammatory bowel illness attributed to a combination of genetic, immunologic, and environmental factors. It usually occurs gradually, although it can occasionally start abruptly. Two age peaks in occurrence are described: one in early adulthood and the other in patients after 60.[7] In North America, CD is estimated to affect 3.1–14.6 people/100,000 person-years. The greatest prevalence was recorded from a study in a pediatric age range, at 48%. The prevalence percentage is expected to be between 50% and 20%.[8] Although the specific causes of CD are unknown, several studies suggest that immune system alterations and exposure to environmental risk factors, such as gastrointestinal bacteria, may induce the condition. Dysregulation of immune system components in the gut may be caused by increased local proinflammatory cytokine production and deficiencies in counter-regulatory mechanisms.[7] In the majority of

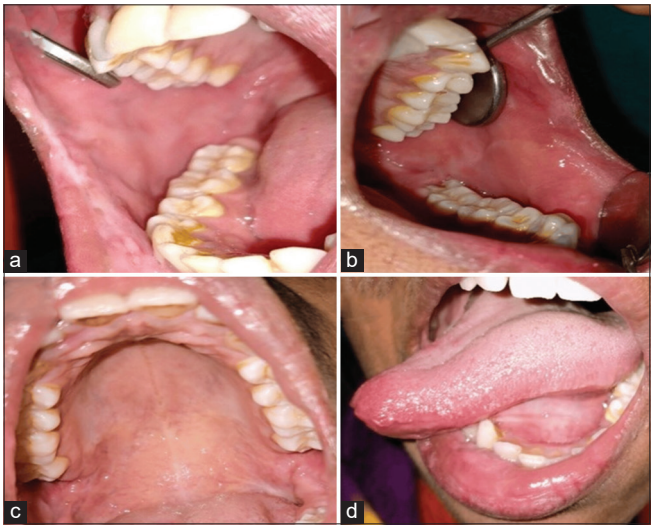

Figure 4: Post-operative evaluation after 3 weeks revealed complete closure of the oral lesions

instances, intestine involvement occurs before the oral lesion. In 5–10% of affected patients, oral lesions may be the first indicator, followed by gastrointestinal problems.[9] Oral lesions in CD typically appear on labial mucosa, buccal mucosa, vestibular sulcus, and gingiva. In around 25% of instances, mucogingivitis is present, followed by persistent superficial oral ulcers similar to small aphthous ulcers in 8% of cases. Approximately 6% of individuals had persistent lip edema, cobblestone papules of the buccal mucosa, and vestibule.[9]

The medical history and clinical examination results serve to guide the research. The biopsy is a critical step in determining the accurate diagnosis; granulomas found during histological examination are common in both OFG and Oral CD (OCD). The only approach to rule out CD is by clinical presentation. OFG may eventually lead to the diagnosis of CD. Recently, it was revealed that four out of six children with OFG in early childhood developed CD during follow-up.[10] OCD therapy calls for a multidisciplinary approach and thorough consideration of available therapeutic options. To separate OFG from other disorders such as infections or systemic diseases, a precise diagnosis is necessary, which may involve a clinical examination and maybe a biopsy. To control localized lesions and minimize inflammation, topical corticosteroids, such as triamcinolone acetonide, are frequently the first step in pharmacological therapies. Systemic corticosteroids, such as prednisone, can be used for broad lesions, while intralesional steroid injections may be utilized for more severe instances. When treating chronic instances that may not respond to existing medications, immunosuppressants such as methotrexate or azathioprine may be an option. Infliximab, a biological medicine, may be recommended for severe symptoms, particularly when linked to CD. Importantly, patients may require enteral nutrition or dietary guidance since they are uncomfortable when eating, which can result in dietary limitations or deficits. Surgery may be a possibility for non-responsive lesions, and maintaining good oral hygiene is important in preventing subsequent infections.

Effective management of OFG requires a multidisciplinary approach involving collaboration between mental health specialists, dietitians, gastroenterologists, dentists, and dermatologists. A coordinated approach to patient care is ensured by constant communication among the team members, including exchanging information on treatment outcomes and problems. Encouraging patients to actively engage in their care by providing them with information about their disease and available treatments is essential for encouraging them to follow dietary and pharmaceutical recommendations. The emotional difficulties brought on by a chronic illness are addressed by offering psychosocial help through therapy or support groups. To assess the effectiveness of treatment and modify therapy as appropriate, follow-up consultations are crucial. Creating referral networks improves treatment by enabling personalized dietary guidance or mental health assistance. Ultimately, complete management is ensured by developing an integrated care plan that addresses the patient's dental, gastrointestinal, and psychological health. This improves the patient's overall quality of life and results.

CONCLUSION

The following case highlights the difficulty in identifying and managing CD, particularly in the presence of oral symptoms. A comprehensive diagnostic approach is necessary to distinguish CD from other illnesses, as evidenced by the patient's symptoms, which include systemic difficulties and recurrent mouth ulcers. Anti-inflammatory medications, immunosuppressants, and biologics must be used in conjunction for effective treatment; systemic therapy should be the main emphasis to address oral and intestinal symptoms. This instance highlights the significance of controlling CD using an all-encompassing, multidisciplinary approach and customizing treatment to get the best results.

REFERENCES

1. Mays JW, Sarmadi M, Moutsopoulos NM. Oral manifestations of systemic autoimmune and inflammatory diseases: Diagnosis and clinical management. J Evid Based Dent Pract 2012;12:265-82.
2. Baumgart DC, Carding SR. Inflammatory bowel disease: Cause and immunobiology. Lancet 2007;369:1627-40.
3. Abraham C, Cho JH. Inflammatory bowel disease. N Engl J Med 2009;361:2066-78.
4. Boirivant M, Cossu A. Inflammatory bowel disease. Oral Dis 2012;18:1-15.
5. Boraz RA. Oral manifestations of Crohn's disease: Update of the literature and report of case. ASDC J Dent Child 1988;55:72-4.
6. Hyams JS. Extraintestinal manifestations of inflammatory bowel disease in children. J Pediatr Gastroenterol Nutr 1994;19:7-21.
7. Pittock S, Drumm B, Fleming P, McDermott M, Imrie C, Flint S, *et al*. The oral cavity in Crohn's disease. J Pediatr 2001;138:767-71.
8. Kim SK, Lee ES. Orofacial granulomatosis associated with Crohn's disease. Ann Dermatol 2010;22:203-5.
9. Atarbashi-Moghadam S, Lotfi A, Atarbashi-Moghadam F. Pyostomatitisvegetans: A clue for diagnosis of silent Crohn's disease. J Clin Diagn Res 2016;10:ZD12-3.
10. Eckel A, Lee D, Deutsch G, Maxin A, Oda D. Oral manifestations as the first presenting sign of Crohn's disease in a pediatric patient. J Clin Exp Dent 2017;9:e934-8.

Source of Support: Nil. **Conflicts of Interest:** None declared.