

Case Series

How Oral Specialists Can Help Diagnose and Manage Extra-Digestive Inflammatory Bowel Disease Complications

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Keywords

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Abstract

Identification of extra-digestive manifestations of inflammatory bowel disease (IBD) is essential. The oral cavity is a preferential site in which gingival enlargement may be one of these manifestations. We present, in this article, two original cases and a concept map that highlights the need for a close collaboration between the dental surgeon or oral specialist, the dermatologist, and the gastroenterologist. In the first case, the strictly local management of a systemic IBD oral complication, can relieve and answer the patient's complaint without modifying or disrupting the systemic treatment already implemented by the gastroenterologists. In the second case, the dental surgeon's diagnosis of gingival enlargement turns out to be the inaugural manifestation of Crohn's disease and allows early treatment of the intestinal pathology.

These two cases illustrate the close link between the oral cavity and IBD. Knowledge and multidisciplinary management of these manifestations such as proposed in the concept map are essential for clinicians for the early diagnosis and the improvement of the oral and general quality of life of patients suffering from IBD.

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Introduction

Gingival enlargement is a common oral manifestation that has multiple aetiologies among them poor dental plaque control, hormones, medications, and chronic systemic diseases [1]. It can also be associated with pain, ulcerations, tooth migration and mobility, chewing and speech disorders and have unattractive, aesthetic consequences. This can highly impair the patient's well-being and alter their quality of life. Also, identifying the aetiology and establishing the correct diagnosis allows setting up an appropriate therapy and limits the risk of recurrence. Patients with chronic inflammatory bowel disease (IBD), Crohn's disease being the most common, can be subject to extra-digestive, oral, manifestations. These manifestations are generally polymorphic: aphthous, gingival hypertrophy, cobblestone patterns, ulcerations but also pyostomatitis vegetans and cheilitis [2]. They should however not be neglected because they can be either associated with the pathology itself or the consequences of its treatment. They can also be inaugural of the disease or reflect a relapse of the disease at the time of treatment.

We report here through two different clinical cases, the course of care and strictly oral management of such gingival enlargements and propose a concept map as a clinician's guide.

Case Report

Patient 1, a 42-year-old female, was referred to us by the gastroenterologists for intense gingival pain and cheilitis evolving for a few weeks (Fig. 1a, b). She was under treatment for Crohn's disease by an immunomodulatory anti-TNF α drug, infliximab (REMICADE®), since 2001.

Patient 2, a 25-year-old male, was referred to us by the orthodontists for a recurring gingival enlargement associated with severe tooth migration evolving for several years (Fig. 1c). He had a family history of Crohn's disease. Oral and facial clinical exams also showed signs of labial hypertrophy with cheilitis and a cobblestone type of hypertrophy on the inner side of the cheeks (Fig. 1d).

Gingival biopsies were performed on both patients. Anatomopathological examination revealed a squamous papillomatous epithelium without cytonuclear atypia resting on an essentially lympho-plasmocytic inflammatory chorion. Giganto-cellular granulomas were found in both cases, without the presence of caseous necrosis. These typical forms of granulomas can be either isolated in the oral cavity or found in cases of systemic diseases such as IBD (Fig. 2).

After having the stability of her Crohn's disease reassessed by the gastroenterologist, topical dermocorticoids (Clobetasol, Dermoval®) were prescribed for patient 1. After 15 days of bi-daily application, complete clinical and symptomatic improvement was observed (Fig. 3a).

For patient 2, a complete endoscopic examination was carried out, revealing no signs of IBD lesions in the digestive tract. A local gingivectomy was carried out in order to remove the

gingival overgrowth. Also, given the overall general clinical picture, this oral manifestation being considered as inaugural of the pathology, a preventive treatment with adalimumab (HUMIRA®) was introduced after a multidisciplinary meeting. No gingival recurrences were observed during the 18-month follow-up (Fig. 3b). However, an annual follow-up of the digestive tract was set up in order to monitor the appearance of intestinal symptoms since they can appear several years after the oral lesions.

Discussion

IBDs are characterized by chronic intestinal inflammation evolving into a variable course of flare-ups and remission [3]. Although the aetiology is still imperfectly known today, the main hypothesis is an inadequate intestinal immune response to bacteria in the commensal flora, probably triggered by environmental factors associated with genetic predisposition. The most common symptoms are pain, abdominal cramps, diarrhoea, rectal bleeding, weight loss, faintness, tiredness. 50% of IBD patients develop oral lesions with a wide variety of clinical forms such as linear fissures, white reticular tags, cobblestone mucosal patterns (pathognomonic of Crohn's disease), gingival enlargement and labial swelling [2, 4]. They may be specific to the intestinal pathology (inaugural, at the same time or after the appearance of gastrointestinal symptoms) or reactive, non-specific consequences of drug treatments and nutritional deficiencies due to malabsorption and malnutrition [5]. They may also be early signs of therapeutic escape or disease relapse. Management of these patients is multidisciplinary, it requires a close collaboration between dental surgeons or oral specialists, dermatologists, and gastroenterologists. For this, we propose a clinician's guide in the form of a concept map (Fig. 4). In a known IBD context, the appearance of oral lesions should lead to address to gastroenterologists to reassess the digestive pathology. The oral biopsy will confirm if the lesion is related to the digestive pathology or its treatment. If so, treatment of the oral manifestation will start with the application of topical steroids such as clobetasol (Dermoval®) several times a day for at least 15 days. If insufficient, systemic steroids may be used at a dose of 0.8 mg/kg/day for 1 month [6]. In addition, in some cases, local surgical management by gingivectomy is necessary since gingival overgrowth renders oral hygiene difficult, aggravating the oral manifestation. In most cases, complementary examinations (other biopsies, biological analyses, radiological examinations, digestive tract extension check-ups) are to be carried out in order to eliminate differential diagnoses. Indeed, there are many chronic pathologies with granulomatous lesions that may have as an oral expression gingival enlargement (Wegener's disease, sarcoidosis, orofacial granulomatosis, tuberculosis, or fungal infections) [7–10]. Establishing the correct diagnosis represents a major impact in the management of not only IBD but also the oral lesions. Close collaboration between oral specialists, dermatologists, and gastroenterologists is essential in order to respond to patients' complaints, prevent non-aggravation of the intestinal state, and improve overall quality of life.

Statement of Ethics

All patients in this study have given their written informed consent to publish their case (including publication of images).

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

Charlotte Thomas and Mylène Dimmock contributed substantially to the design of the work and to drafting the work.

Cyrielle Gilletta contributed substantially to the interpretation of the data of the work and revised it critically from the gastroenterological point of view.

Béatrice Barres contributed to the acquisition and analysis of the anatomopathological samples and revised the work accordingly.

Carle Paul and Sarah Cousty contributed substantially to the interpretation of the data of the work and revised it critically from the dermatological point of view.

Sara Laurencin-Dalicieux contributed substantially to the conception and design of the work, interpretation of the data, and drafting of the manuscript.

All authors approved of the final version of this case report and agree to be accountable for all aspects of the work.

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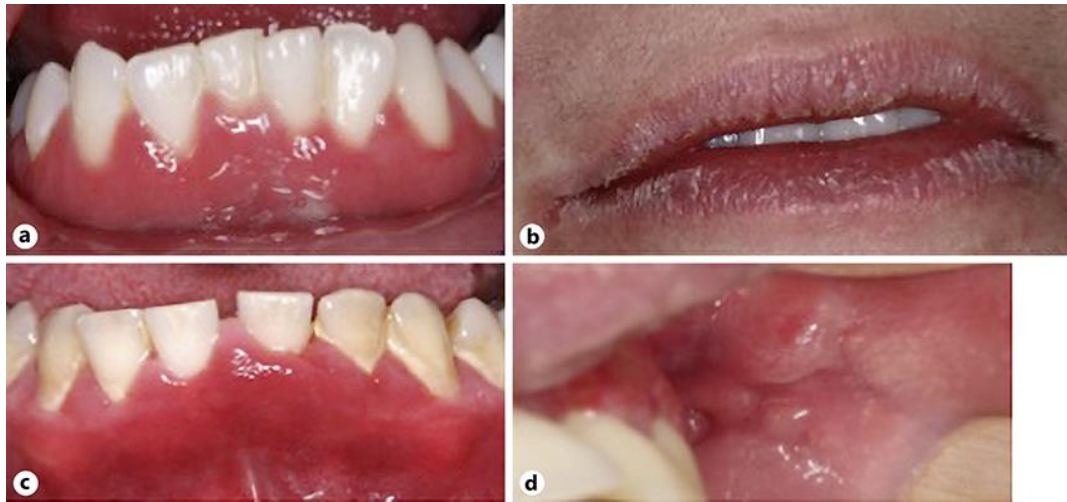


Fig. 1. a Gingival enlargement in patient 1. b Cheilitis in patient 1. c Gingival enlargement in patient 2. d Cobblestone lesion on the inner side of the cheek in patient 2.

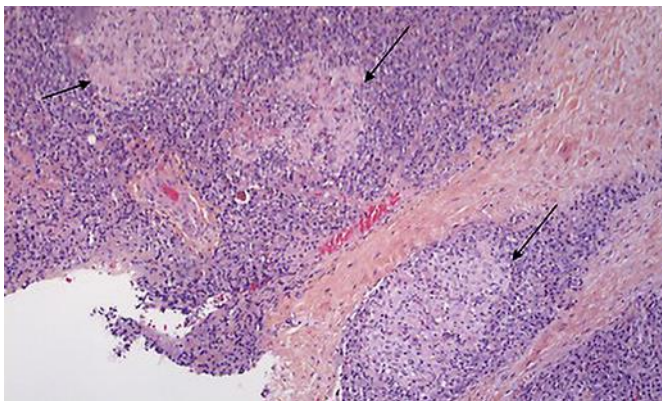


Fig. 2. Histopathological sections of gingival biopsies stained with haematoxylin-eosin revealing the presence of granulomas ($\times 10$).



Fig. 3. a Clinical situation after 15 days of local topical corticosteroid treatment (patient 1). b clinical situation, 18 months after surgical treatment by gingivectomy (patient 2).

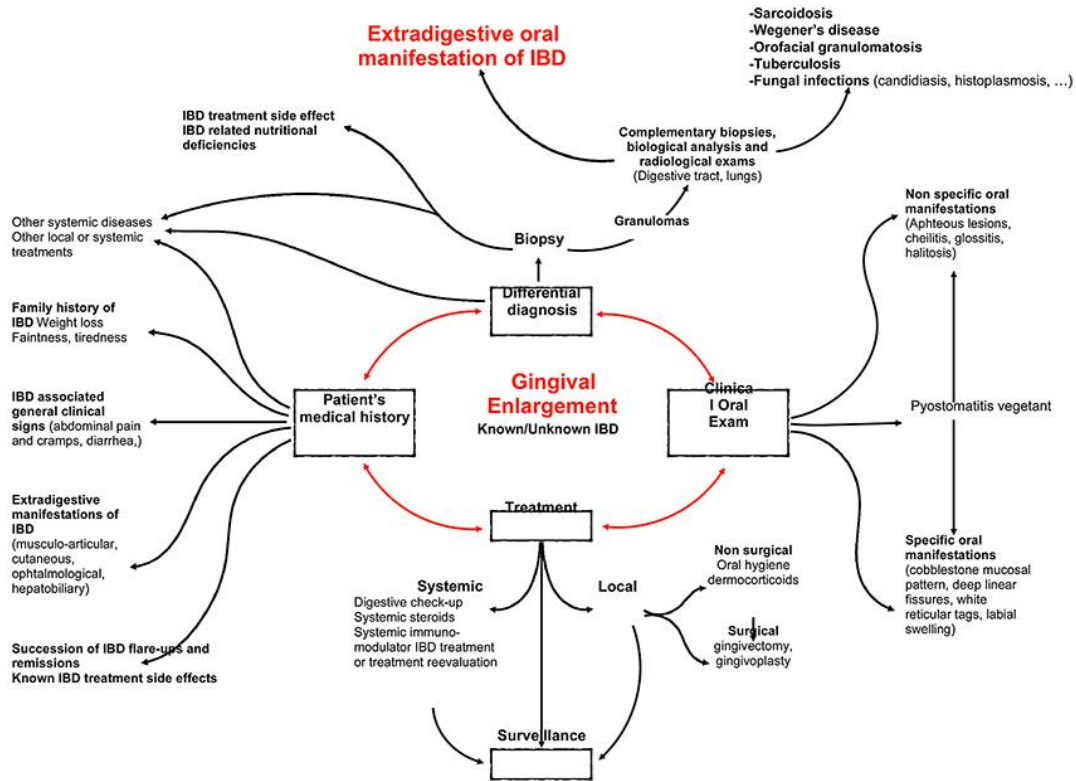


Fig. 4. Gingival enlargement and IBD concept map and clinician's guide.