

CASE REPORT

Gingival manifestation of Crohn's disease in a paediatric patient: A case report

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Funding information

Open Access Funding provided by Universita degli Studi di Padova
Blended DEAL: CARE

Abstract

Crohn's disease (CD) is a complex multisystemic inflammatory disorder and part of the inflammatory bowel diseases (IBDs). About 20% of the subjects affected by IBDs are paediatric patients and 21% of them present extraintestinal manifestations. Among the extraintestinal manifestations of CD, oral manifestations are rather frequent (20–50% of affected people) and can be difficult to identify, being frequently aspecific and not pathognomonic. Their early diagnosis by the dentist is extremely important, because extraintestinal manifestations can precede intestinal symptoms, in all patients but especially in children. The aim of this paper is to report the case of a CD adolescent patient that presented a gingival manifestation.

KEYWORDS

angular cheilitis, gingival inflammation, inflammatory bowel disease, oral crohn, paediatric crohn

INTRODUCTION

Crohn's disease (CD) is one of the main inflammatory bowel diseases (IBDs), which are characterized by idiopathic and chronic inflammatory lesions of the gastro-intestinal tract.¹ In North America and Europe, over 1.5–2 million people suffer from IBD; in particular, from 1990 to 2016,² the prevalence of CD in these countries has been of 135.6/100.000 inhabitants. Traditionally, IBDs are linked with western countries, where they are a cause of morbidity, mortality, and substantial costs for public health.² Only in the last decades, IBDs are spreading in Asia, South America and Africa.³

Crohn's disease is a chronic inflammation of, potentially, the entire gastro-intestinal tract, from mouth to anus.³ Bowel inflammation is triggered in genetically predisposed individuals by dysregulated immune response. Several immune, genetic and environmental factors seem to be related with CD.³ There are also some correlations with lifestyle and industrialization.

Children and adolescents also suffer from IBDs: indeed, around 20–25% of the people affected by IBDs are younger

than 18 years old. In particular, the incidence of CD in the paediatric population is increasing from 2.5 to 11.4 per 100.000, with a prevalence of 58 per 100.000.⁴

No pathognomonic markers are associated with CD, so diagnosis includes considerations about family's medical history, clinical manifestations, serological and endoscopic exams. Clinical symptoms include abdominal pain, diarrhoea, weight loss and extra-intestinal manifestations, such as primarily peripheral arthritis, ankylosing spondylitis, uveitis and erythema nodosum.³ The location of the intestinal lesions in paediatric patients is combined ileo-colonic disease in 53%, isolated colonic in 27% and limited cecal in 16%.⁴ According to Paris classification, there are four main features to be considered when evaluating CD in a paediatric patient: age of onset, disease location, disease behaviour and effects on the patient's growth.⁵ Therapy for CD is mainly pharmacological, through corticosteroids and immunomodulatory agents; surgery is not curative, and it is used only for complications such as perianal fistulas or abscesses and mechanical impediment of the intestinal contents flow.⁴

[Correction added on 09-May-2022, after first online publication: CRUI funding statement has been added.]

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Extra-intestinal manifestations (EIMs), such as growth and development impairment and perianal disease, occur in 21% of children with IBDs. These non-classical manifestations are often the first signs of IBD. It is calculated that, for this reason, there would be a delay in diagnosis of paediatric CD of 3.4 months.⁶

Oral manifestations (OMs) are part of EIMs of CD. Their prevalence in CD patients is 20–50% of the affected people. Lesions include ulcers, papules and edema of the lips, gingiva and vestibular sulci. In children, aphthous stomatitis occurs in 10–46%.⁷ Other authors report persistent lip swelling, cobble stoning of the oral mucosa, mucogingivitis, deep linear or serpiginous ulcerations surrounded by epithelial hyperplasia, tissue tags or polyps.⁸ Jajam et al.⁷ classified them into specific oral lesions (labial swelling, cobble stoning of mucosa, mucogingivitis and linear ulcerations) and non-specific oral lesions (aphthous stomatitis, angular cheilitis and pyostomatitis vegetans). OMs can present before or after the diagnosis has been made, but, especially in paediatric patients, they usually precede or coincide with intestinal symptoms, so CD might be considered for paediatric patients with the above-reported oral lesions.⁹

Recognizing OMs, and EIMs in general, could represent a valuable tool to reduce the delay of CD's diagnosis in paediatric patients. Despite that consideration, in the literature, there are only few case reports about CD's OMs and few reviews about oral lesions in all IBDs.

Therefore, the purpose of this paper is to report an oral manifestation of CD in a paediatric patient.

DESCRIPTION OF THE CASE

In November 2020, a 15-year-old girl was referred to our group of Oral Pathology by the department of Paediatric Gastroenterology of Padua University's Hospital.

The patient was diagnosed with "Crohn's disease with pancolitis" in March 2018 and had already undergone several hospitalizations, due to a long history of perianal complications (the first one occurred in 2012). She was placed an ileostomy in March 2018 and had been treated with monthly infusions of Infliximab (IFX, a TNF-inhibitor) since April 2018. This therapy had been suspended only once, in March 2019, because of the appearance of labial herpetic lesions. The patient also suffered from allergic asthma, with respiratory attacks becoming less frequent and lighter.

At the time she came to our clinic, she was being treated with Adalimumab (ADA, a different TNF-inhibitor) and Azathioprine (an immunosuppressant) and the gastrointestinal disease was under control. She was also taking a vitamin D supplement and Levocetirizine (an antihistamine, to be taken when needed), while she was no longer being treated with aerosol of Beclomethasone (a steroid anti-inflammatory medication), since her allergic asthma was improving.

The patient was referred to our attention by her gastroenterologists because of some erythematous intraoral lesions, which they wanted to make sure were OMs of CD.

CLINICAL RELEVANCE

Scientific Rationale for the Study: To highlight this gingival manifestation in a paediatric patient affected by Crohn's disease and how the final diagnosis of non-pathognomonic oral lesions should be based on the correlation between the clinical aspect and histopathological features.

Principal Findings: Gingival reddening and swelling are not so frequently reported in the literature as an EIM of Crohn's disease (as is, for example, angular cheilitis) and could be easily mistaken with gingival inflammation of another nature.

Practical Implications: The authors suggest that clinicians should always consider, in the process of differential diagnosis, that oral lesions could be manifestations of a systemic disease.



FIGURE 1 Unusual gingival presentation of CD

At the clinical examination, the gingiva and alveolar mucosa, especially in the upper arch, appeared reddened and edematous. (Figure 1) They were not painful, and the patient did not report any particular symptomatology. Angular cheilitis was also present on the left labial commissure. (Figure 2).

No other OMs of CD, such as mucosal tags, ulcers or cobblestoning, were present. The patient showed an ideal condition of oral hygiene, without presence of plaque nor signs of periodontal disease.

We suggested the execution of a histological examination of the lesions, in order to perform a differential diagnosis between an intraoral manifestation of CD and other orofacial granulomatous diseases (such as granulomatosis with polyangiitis, the once called Wegener's granulomatosis) or sarcoidosis.

Under local anaesthesia, a circular sample (5mm diameter) of the buccal alveolar mucosa in the upper right arch was collected through a punch biopsy.



FIGURE 2 Angular cheilitis

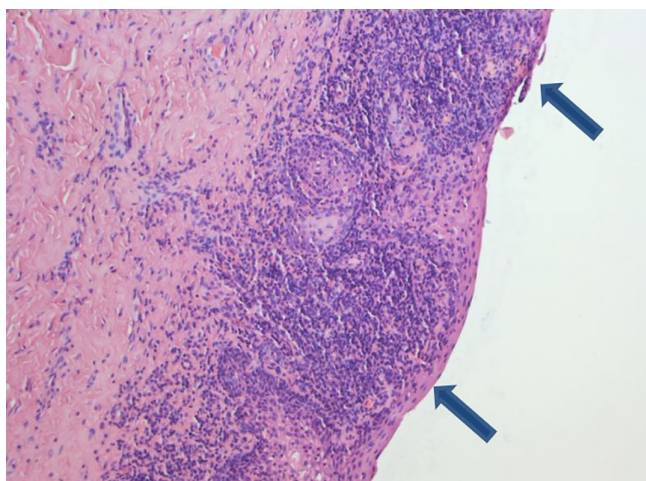


FIGURE 3 Histological examination: evidence of epithelial atrophy and ulceration (arrows). Magnification 10x

The histological examination resulted in “fragment of oral mucosa with lymphocyte and monocyte inflammatory infiltration of the submucosa and the squamous epithelium, with formation of granulomatous-like aggregates within the submucosa and the deeper derma. Epithelial ulceration associated with leucocyte infiltration. Gigantic cells were not found. The immunohistochemical characterization found the prevalence of T lymphocytes (CD3 = 70%), B lymphocytes (CD20 = 20%) and macrophages (CD68 = 10%).” (Figures 3–4).

The final diagnosis established that the histological findings were compatible with intraoral manifestations of CD.

DISCUSSION

CD is a complex multisystemic inflammatory disorder in which genetics, microbiota and immune response interact in a complex and unpredictable way. This interaction can produce intestinal and extra-intestinal manifestations, where the second type is often prior or contemporaneous with intestinal symptoms.⁸

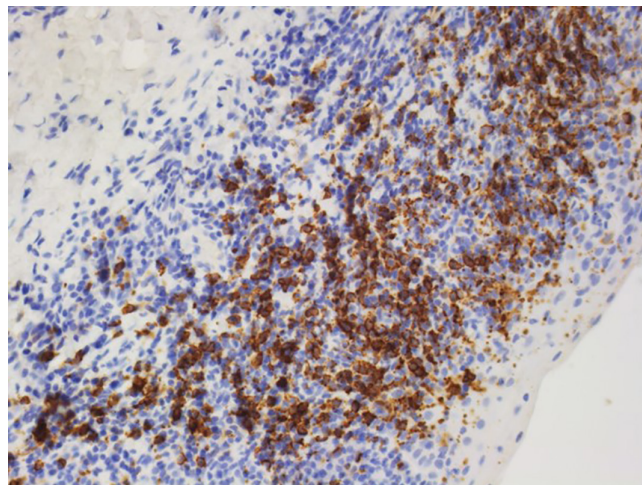


FIGURE 4 Immunohistochemistry analysis: Inflammatory cells phenotyping with antibody against CD20+ B-lymphocytes. Magnification 20x

In paediatric patients, there is a higher prevalence of OMs, especially in severe phenotypes.⁹ Lesions can be painful and are associated with higher risk for tooth decay, since it is difficult for symptomatic patients to maintain a correct oral hygiene. This occurrence could compromise children's oral function and therefore their already impaired quality of life. Short-term therapies with topical corticosteroids for painful lesions and oral hygiene are the only way to treat this pattern.⁹

Oral lesions are often non-specific, such as forms of stomatitis, angular cheilitis and glossitis.⁸ Most of them could challenge paediatric dentists because they are similar to other orofacial granulomatosis (OFGs) manifestations. OFGs are idiopathic chronic inflammations of the oral mucosa and lips.¹⁰ There is a non-well-established connection between OFGs and systemic diseases like CD or sarcoidosis. Evidence is not enough to establish whether OFGs are always specific entities.¹⁰ In certain cases, indeed, an OFG can be present together with a systemic disease.¹¹

In our case, we discuss oral lesions in a patient affected by CD, lesions that could also be related to other pathologic entities. Since our patient showed a perfect oral hygiene, with no plaque accumulation nor bleeding on probing, and undergoes periodic dental examinations, we excluded infective/periodontal causes of gingival inflammation and focused on oral manifestations of systemic diseases. Discolouration and swelling of alveolar mucosa can be, indeed, OMs of CD⁷ and enter differential diagnosis with manifestations of many other systemic diseases, such as ulcerative colitis, granulomatosis with polyangiitis, leukaemia, celiac disease, gastroesophageal reflux and Plummer-Vinson syndrome.

Being OMs of systemic disease frequently aspecific, diagnosis is based on the correlation between the clinical aspect and histopathological features; therefore, a biopsy of the lesions is necessary.¹²

In our case, the patient is orally asymptomatic, so, after diagnosis, we decided not to treat her and keep her monitored, to assure the maintenance of an optimal oral hygiene, which was fundamental to exclude plaque-induced gingivitis from differential diagnosis.

Maintaining a perfect oral hygiene appeared essential also because our patient requested an orthodontic treatment. In the literature, there is only one paper regarding orthodontic treatment in CD patients,¹³ where the authors affirm that presence of condylar arthritis or lesions involving the periodontium represents a contraindication for orthodontic treatment and ultimately decided not to treat their patient. Our patient, however, shows no signs of arthritis or periodontal disease, is asymptomatic and keeps an optimal oral hygiene, so, in our opinion, the orthodontic treatment is not contraindicated. We suggested an orthodontic treatment with aligners, to minimize plaque retention and facilitate the maintenance of an optimal oral hygiene.

The decision to report a case of gingival presentation of CD was made to suggest that dentists, and especially pedodontists, can play an essential role for the early diagnosis of CD (and many other systemic diseases¹⁴) and can help patients and other specialists manage this complex chronic pathology through time.

ACKNOWLEDGEMENT

Open Access Funding provided by Università degli Studi di Padova within the CRUI-CARE agreement.

CONFLICT OF INTEREST

No conflict of interest has been declared by the authors.

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How to cite this article: Giaccaglia F, Tomasin M, Angelini A, Bacci C. Gingival manifestation of Crohn's disease in a paediatric patient: A case report. *Oral Surg*. 2022;15:573–576. <https://doi.org/10.1111/ors.12682>