Pyostomatitis vegetans leading to Crohn's disease diagnosis

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Pyostomatitis vegetans (PV) is a rare oral condition of unknown pathogenesis, usually considered a specific marker of inflammatory bowel disease (IBD), in particular Crohn's disease [1-5]. PV manifests with yellowish, linear pustules set on an erythematous base that are usually called "snail track ulcerations" [3,4]. Hallopeau first introduce the term PV for similar skin lesions in 1898, and McCarthy described the oral analogue as PV in 1949 [6]. The presentation of PV may coincide, precede or follow intestinal involvement [6,7].

We present a rare case of PV that led to the diagnosis of Crohn's disease, in order to emphasize the association of oral and gastrointestinal (GI) diseases.

A 58-year-old man was referred for evaluation of "white lesions that looked like pus" on the labial gingiva. The lesions peeled off during tooth brushing, without causing any discomfort. They had been initially diagnosed as pseudomembranous candidiassis and unsuccessfully medicated with Miconazole nitrate oral gel. The patient's medical history disclosed that during the last few months he had been suffering from bloody diarrhea. Complete colonoscopy was performed by a GI specialist, and a diagnosis of "hemorrhoids" was given.

Clinical examination revealed numerous yellow-white mucosal pustules on the labial maxillary and mandibular gingiva, as well as the hard palate. They were asymptomatic, set on an erythematous base, had the characteristic appearance of "snail-tracks", and easily peeled off upon rubbing with gauze (Fig. 1). The rest of the mucosal examination as well as head and neck examinations were within normal limits. The patient did not report the presence of any skin lesions.

A recent complete blood count, performed for a regular check-up, showed anemia [RBC 4.28 (4.50-6.30* 10^6 /µL), HB 10.7 (14-18 g /100 mL), HCT 34.8 (40-52%), MCH 25.0 (26-32 pg), MCHC 30.7 (32-36 g /100 mL), ferritin 5.20 (25-380 g /100 mL), Fe 44 (65-157 mg/dL), peripheral eosinophilia, EOS 15.6 (1-3%), and increased erythrocyte sedimentation rate 38 (<20 mm/h)].

With the clinical diagnosis of PV, a partial biopsy was performed under local anesthesia. Microscopic examination of 5 μ m thick formalin-fixed and paraffin-embedded tissue sections stained with hematoxylin and eosin, showed acanthosis and pseudoepitheliomatoid hyperplasia of the focally detached parakeratinized stratified squamous epithelium, and



Figure 1 "Snail-track" mucosal pustules on an erythematous base on maxillary gingiva

intraepithelial and subepithelial microabscesses, composed of eosinophil and polymorphonuclear cells. The underlying connective tissue demonstrated a mixed inflammatory infiltrate, consisting of eosinophils, neutrophils and lymphocytes. No *Candida* hyphae were identified in a PAS stained section.

With the provisional diagnosis of IBD, the patient was referred to a GI specialist. A new complete colonoscopy revealed ulcerative-type pancolitis. Biopsies revealed transmural pattern of inflammation and were interpreted as "consistent with Crohn's disease".

The oral lesions persisted 2 months after systemic medication with Mesalazine 500 mg daily, probably due to the low dose of medication. The patient was instructed to rinse his mouth three times daily with chlorhexidine 0.12%, but he was lost to follow up.

In conclusion, PV may be rare, but its association with IBD necessitates its prompt recognition. Thorough evaluation of the patient, in particular when GI symptoms and signs are subtle and may be easily overlooked, is very important, as this could result in early diagnosis of the underlying disease.

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Conflict of Interest: None

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Received 14 October 2012; accepted 5 November 2012