Generalized Gingival Hyperplasia and Rectorrhagia in a 13-Year-Old Boy

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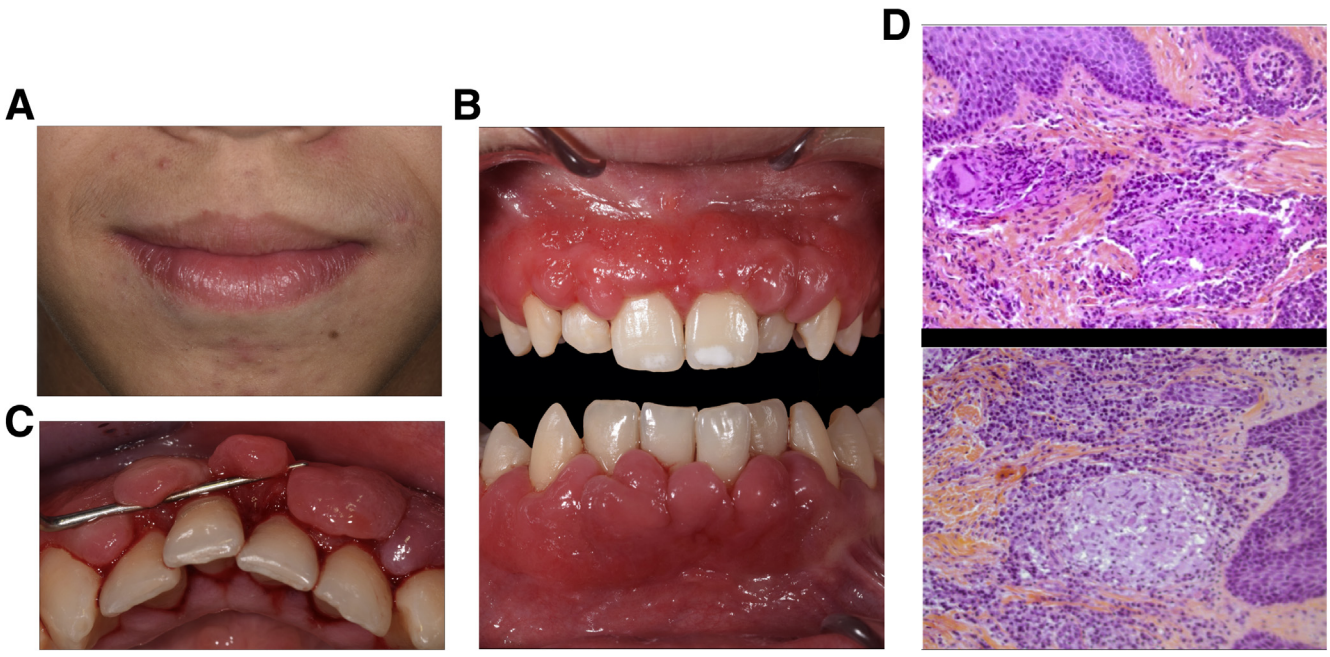

Question: A 13-year-old boy of Arab origin was referred to the Department of Periodontology with a chief complaint of generalized bleeding gums for 3 years, which had an impact on his daily quality of life. The young patient did not report any particular symptoms other than a chronic fatigue that hampered his sport activities. The patient's mother mentioned 2 episodes of perianal abscess that were surgically drained at the age of 6 months and 1 year and few episodes of rectorrhagia during childhood. She also noticed a year ago, swollen lips and mouth breathing.

The patient's body mass index is 21.5 kg/m^2. The extraoral examination showed an asymmetric macrocheilia and mouth breathing (Figure *A*). Periodontal examination highlighted a generalized gingival hyperplasia mainly in the vestibular area extending beyond the mucogingival junction and covering a part of the clinical tooth crown (Figures *B*, *C*). The gingiva presented a granulomatous aspect associated with a dentomaxillary discrepancy. The plaque index and bleeding on probing index (estimating gingival inflammation) were 42% and 48%, respectively. Previous sessions of scaling were performed once a year by a general dentist, without any observable impact on the gingival aspect.

Blood samples and gingival biopsies were taken. Normocytic nonregenerative anemia was diagnosed associated with an iron deficiency. Gingival biopsies revealed the presence of a parakeratinized epithelium with hyperplastic areas. The inflammatory infiltrate was rich in plasma cells, noncaseating granulomas, and epithelioid cells (Figure *D*). The patient was then referred to the Pediatric Pulmonology and Gastroenterology Departments for further workup. Esophagogastroduodenoscopy and colonoscopy were performed with biopsy at the level of the esophagus, stomach, duodenum and colon, showing several inflammatory areas associated with numerous ulcerations.

What is the most likely diagnosis?

Look on page 2032 for the answer and see the *Gastroenterology* web site (www.gastrojournal.org) for more information on submitting your favorite image to Clinical Challenges and Images in GI.

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Conflicts of interest
The authors disclose no conflicts.

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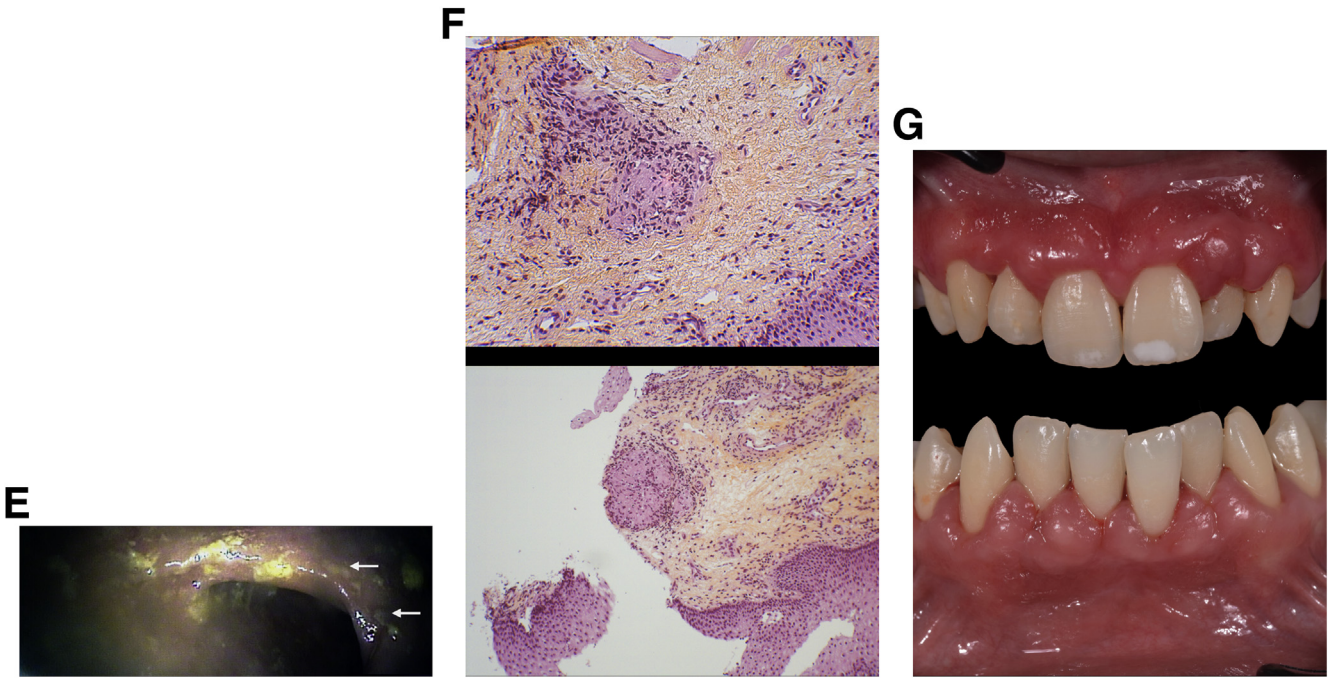
Answer to: Image 2 (Page 2030): Crohn's Disease



Melkersson-Rosenthal syndrome was excluded because of the lack of clinical signs. Absence of exophytic pustules on the oral mucosa ruled out pyostomatitis vegetans. Pulmonary radiologic examination (computed tomography scan) excluded sarcoidosis and tuberculosis. Biological tests for granulomatosis with polyangiitis proved positive for cytoplasmic antineutrophil cytoplasmic antibodies and negative for antisaccharomyces cerevisiae antibodies. The esophagogastroduodenoscopy and colonoscopy reports were conclusive, leading to the diagnosis of Crohn's disease (CD) localized at the esophagus, stomach, and colon and associated with gingival hyperplasia. Aphthoid ulcers were mainly observed in the transverse and sigmoid colons (Figure *E*).

The histopathological analyses of the gastro-intestinal biopsies showed the presence of noncaseating granulomas and epithelioid cells, with plasma cells and neutrophils in the inflammatory infiltrate (Figure *F*). After discussion in a multidisciplinary meeting, immunosuppressive (azathioprine) and corticosteroid agents were prescribed associated with supplementations in vitamins B_{12} and B_9 and iron to correct deficiencies. Simultaneously, periodontal treatment was performed by a specialized periodontist including, oral hygiene instruction with scaling and root planing.

At the 3-month follow-up, a partial regression of CD is observed together with a significant reduction of the gingival hyperplasia and inflammation (Figure *G*). The patient was shifted to a biological therapy with infliximab. Gingivectomy will be eventually carried out to further decrease the gingival enlargement and promote optimal long-term plaque control, if complete remission is not observed. A close monitoring of both the periodontal and intestinal manifestations is needed. Extraintestinal manifestations of CD (eg, arthritis, osteoporosis, and oral diseases) are common, especially in pediatric patients, and may be present several years before CD diagnosis.[1] It is estimated that up to 50% of CD patients present with oral manifestations.[2]

Patients with CD are at increased risk of having impaired oral health and particularly periodontitis (odds ratio, 3.64; 95% confidence interval, 2.33-5.67) compared with patients without CD.[3] This susceptibility can be explained by the similarities in the pathogenicity between periodontitis and CD, particularly related to the inflammatory pathways and dysbiotic mechanisms in the microbial environment. This case report highlights the key role of dentists in the screening and diagnosis of systemic diseases that might present with oral manifestations, and the necessity of a close collaboration between oral health specialists and physicians.

Keywords: Crohn's disease; Periodontal disease; Extra-oral manifestations.

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