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Crohn's disease is a chronic granulomatous inflammatory bowel disease characterized by discontinuous chronic inflammation usually associated with noncaseating granulomas. It may affect any part of the gastrointestinal tract. Oral manifestations of Crohn's disease are well known, and pharyngolaryngeal localizations have been described. Nasal involvement is exceptionally rare. In this article we present a case of nasal involvement in a patient with concomitant acute colitis, subsequently diagnosed as Crohn's disease.

CASE REPORT

A 38-year-old man sought treatment in October 1993 for a severe and bilateral nasal obstruction with watery rhinorrhea and recurrent epistaxis of 10 days' duration. Two weeks before he had noted ulceration of the oral mucosa.

The patient had insulin-dependent diabetes mellitus diagnosed 20 years previously. In 1985 he was investigated for diarrhea with blood and mucus in the stools. A barium enema revealed proctitis. A colonoscopy showed a red, swollen mucosa with multiple erosions within the rectum. The biopsy specimens showed acute and chronic proctitis. The diagnosis of ulcerative proctitis was made. Since 1985 he has had recurrent diarrheas that have been treated with mesalamine with good response. During the 3 months preceding the present disease, he had an exacerbation of diarrhea episodes.

On physical examination he appeared well nourished, afibrile, and not jaundiced. No skin lesions were present. The nasal mucosa was uniformly swollen, completely obstructing the nasal cavity. The left side of the septum was tumefied, and an incision evacuated 1.5 ml of clear fluid.

Fig. 1. Multiple superficial ulcerations were present on the palate, tongue, and oral vestibule (arrows).

Multiple superficial ulcerations were present on the palate, the tongue, and the oral vestibule (Fig. 1). The pharynx and larynx were normal. Laboratory investigations revealed a hemoglobin level of 10.8 gm/dl, an erythrocyte sedimentation rate of 60 mm/hour, and a white blood cell count of 11,300/mm³ (segmented 75%, nonsegmented 3%, eosinophils 7%, basophils 0%, monocytes 6%, and lymphocytes 9%). Chest x-ray film was normal. Because of the suspicion of a septal abscess, the patient was admitted and treated with intravenous clindamycin and gentamicin. In the absence of improvement, a computed tomography scan was performed on the fourth day. It showed diffuse thickening of the entire sinonasal mucosa, particularly of the septum but without a localized collection (Fig. 2). The cultures of the septal drainage were sterile. An immunologic screening was performed. The C-reactive protein level was 13.7 mg/dl (normal, <0.3 mg/dl). Immunoglobulin levels were normal. The complement CH50 level was 158% (normal, 75% to 125%), and the C3 fraction was 1.48 gm/L (normal, 0.63 to 1.35 gm/L). Antinuclear antibody levels were normal. The search for antihuman neutrophil cytoplasm antibody in the serum was negative.
Fig. 2. Axial computed tomography scan shows diffuse thickening of the entire sinonasal mucosa and particularly of the septum. There is also a polypoid aspect and a concha bullosa of the right middle turbinate.

Fig. 3. Nasal mucosa (right middle turbinate): Chronic inflammatory infiltrate between accessory glands with a granuloma (asterisk). Inset, Detail of granuloma. (Hematoxylin and eosin stain; original magnifications ×128 and ×217.)

In the hospital the patient had a new episode of watery diarrhea. The stools revealed the presence of blood and leukocytes, but neither bacterial pathogens (salmonella, shigella, and yersinia) nor parasites. A colonoscopy demonstrated a 20-cm inflamed and stenosed segment of the ascending colon. The mucosa was red, with deep ulcerations. The histologic examination showed chronic granulomatous colitis, which was consistent with Crohn's disease because no pathogens were discovered with special stains (Ziehl-Nielsen, periodic acid–Schiff, Grocott), and cultures and serologies remained negative.

The patient had a direct pharyngolaryngoscopy and anterior rhinoscopy under general anesthesia. The nasal mucosa exhibited severe edema with a polypoid aspect of the right middle turbinate and antrum. Biopsy specimens of the right middle turbinate showed lymphoplasmacytic infiltrate with a small focus of macrophages suggestive of a microgranuloma (Fig. 3). The Ziehl-Nielsen stain on this material was negative. The biopsy specimens of the oral mucosa showed submucosal microabcesses without granulomas. Corticotherapy with prednisone 60 mg/day was introduced with a rapid resolution of the diarrhea and of the nasal obstruction. One month later the patient was asymptomatic, and the nasal mucosa had a normal appearance. He remained asymptomatic during the following 6 months.

DISCUSSION

Thirty-six percent of patients with Crohn's disease had extraintestinal manifestations in a series of 498 patients reviewed by Greenstein et al. In the head
and neck, only oral manifestations were discussed and were found to be present in 4% of the patients. This frequency is similar to that of Croft and Wilkinson, who reviewed 332 cases of Crohn's enteritis and found 20 (6%) cases of oral ulcerations and swollen lips and tongue. Of 100 patients with Crohn's disease, Basu et al. found 9% with oral manifestations. Laryngeal involvement has also been described. The head and neck lesions in patients with Crohn's disease are described as poorly delimited mucosal swellings with ulceration limited to the oral cavity. The pathologic examination usually shows chronic inflammation with rare giant cells and absence of necrosis. True granulomas are rarely seen.

Nasal manifestations seem to be rare since there are only two case reports that evoke this association. In 1985 Kinnear reported a case of chronic atrophic rhinitis with crusting, the biopsy specimens of which showed chronic fissuring and deep ulcerations, with a chronic cell infiltration and granulomatous formation. Ernst et al. reported in 1993 a case of pansinusitis with polyposis in a patient with known Crohn's disease. The biopsy specimens showed ulcers but no granuloma.

In the case presented here, diffuse swelling of the nasal mucosa was associated with polyposis of the middle turbinate. The biopsy specimen revealed chronic inflammation with a focus of macrophages suggestive of granuloma. Several oral ulcerations were also present, the biopsy of which showed microabscesses. These pathologic findings, combined with the absence of evidence for other infections or inflammatory granulomatous diseases, the lack of response to antibiotics and local treatment, and the rapid resolution under systemic corticotherapy, lead us to relate the nasal manifestations to Crohn's disease. In the course of this disease, the severity of the intestinal and systemic manifestation might occult other minor manifestations. Perhaps these patients may not spontaneously report nasal obstruction. This case suggests that the frequency of nasal involvement is probably underestimated in this inflammatory disease.

REFERENCES