A case of ulcerative colitis and pyostomatitis vegetans in an African

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SUMMARY A case of pyostomatitis vegetans and ulcerative colitis in a 26-year-old African is described. The colitis, confirmed clinically and radiologically, antedated the development of pyostomatitis. Systemic corticosteroids apparently cured the pyostomatitis but only caused an improvement in the ulcerative colitis.

The rarity of ulcerative colitis amongst the Nigerian population and of pyostomatitis vegetans as a complication makes the occurrence of these two diseases in a Nigerian an extraordinary event.

Although the prevalence of ulcerative colitis in the Nigerian population is not known with certainty, it is highly probable that the disease is rare. Evans and Acheson (1965) showed that nearly all patients with ulcerative colitis in Oxfordshire, England, eventually reached hospital and could be traced through hospital records. In the experience of the authors, the habits of the urban Nigerian patients with chronic diarrhoea do not differ from their counterparts in Oxfordshire; they soon report to the hospital with chronic complaints of this type. Inspection of the medical records of the Lagos University Teaching Hospital over the years 1962-68 inclusive showed only three patients who had been discharged from the hospital wards with a diagnosis of ulcerative colitis and in whom the diagnosis could be substantiated. Two of these were Europeans.

Case Report

The case to be described here is that of an African patient who was diagnosed as suffering from ulcerative colitis after sigmoidoscopy in 1967. At that time he made a complaint of a sore mouth and intermittent difficulty in swallowing, but no specific oral findings were noted except for gingivitis.

On 19 February 1969 he again attended the Outpatient Department complaining of swollen lips and a sore mouth. At that time the diagnoses of pemphigus vegetans mucosae oris and of chronic candidosis were considered but rejected because of several discrepant features. A thorough search through the literature led to the conclusion that the condition of the patient's mouth fitted exactly the description by McCarthy in 1949 of pyostomatitis vegetans.

The eruption within the oral cavity involved the mucosae of the cheeks, the palate, the buccal and vestibular surfaces of the gums, the lateral margin of the tongue, and the upper and lower lips, just extending into the external surface of the lower lip. There were many slightly raised oedematous, soft, reddish areas which were comparatively well demarcated and studded with small pin-head superficial pustules, and with reddish miliary nodules. There was evidence of increased salivation and the affected areas were only slightly painful to the touch. The teeth were in excellent condition. Figures 1 and 2 show the condition of the lips at the time the lesion was seen on 19 February 1969.

The buccal swabs of the mucosa were examined for *Candida albicans* with negative results. The haemoglobin was 73% and the erythrocyte sedimentation rate 40 mm/hr (Westergren). The patient reported an average of seven loose

Fig. 1 *Lips before treatment.*

Fig. 2 *Everted lower lip before treatment.*

Fig. 4 *Everted lower lip after treatment with prednisone.*

Fig. 3 *Barium enema showing whole colon involved by ulcerative colitis.*

stools daily with blood noticed in them from time to time. His general condition was good. He was afebrile. Sigmoidoscopic examination showed hyperaemia of the rectal mucosa with loss of the normal vascular pattern but there was no bleeding after instrumentation. The stools contained pus cells and red blood cells and the faecal occult blood test was consistently positive. One specimen of stool contained cysts of *Entamoeba histolytica*.

A barium enema showed that the transverse colon, the descending colon, and the sigmoid were involved by ulcerative colitis. The barium-filled contour showed no haustration and finely serrated margins (Fig. 3). There was no shortening of the colon and no undercutting of the mucosal surface, but the post-evacuation mucosal pattern was characteristic of ulcerative colitis.

Unfortunately the piece of tissue biopsied from the inner surface of the lower lip was rather small but the following features were discernible. There were parakeratosis, acanthosis, and spongiosis of the epithelium, and oedema of the papillae of the tunica propria, together with dilatation of the papillary capillaries. The capillary endothelium was swollen, and cellular elements surrounding the capillaries mainly consisted of lymphocytes, histiocytes, eosinophils, and a few neutrophils. Some eosinophils were within the capillary lumen and between the cells of the rete Malpighi.

There were no skin lesions.

A diagnosis of pyostomatitis vegetans and chronic ulcerative colitis was made. Prednisone, 40 mg orally in divided doses, was given daily for seven days and the dose was then decreased to 20 mg daily. Within 48 hours of his starting the treatment a remarkable change took place in the oral mucosa. Swelling subsided, the pinpoint pustular lesions regressed, the rough and granular appearance of the buccal mucosa changed, and hyperaemia lessened. There was less eversion of the lower lip. The photograph in Fig. 4 taken eight days after the start of prednisone therapy shows the improvement. There was also a slight improvement in the bowel condition, with a drop in the number of stools passed each day to four.

During three months' observation after the original treatment, the mouth lesions have remained healed but the colitis remains slightly active with an average of four stools per day. The patient's general condition is good, but as sulphasalazine is not available he is maintained on a small (15 mg /daily) dose of prednisone.

Discussion

Three cases of pyostomatitis vegetans were first described by McCarthy in 1949. Since then only seven other cases have been reported by different authors (Table 1), so far as we can ascertain.

Author	No. of Cases of Pyostomatitis Vegetans	With Pyoderma Gangrenosum	With Ulcerative Colitis
McCarthy (1949)	3	—	2
Margoles and Wenger (1961)	1	1	—
Hays (1961)	1	—	—
Zegarelli and Kutscher (1962)	2	—	2
McCarthy and Shklar (1963)	3	1	3

Table I Cases described in the literature

Five of the later cases were in association with ulcerative colitis. Two of them had pyoderma gangrenosum, a complication of ulcerative colitis which Edwards and Truelove (1964) found in 0.5%, and Goligher, De Dombal, Watts, and Watkinson (1968) in 2% of their large series of patients with ulcerative colitis.

Pyostomatitis vegetans is an even rarer systemic complication of ulcerative colitis, so rare indeed that it is not mentioned as such either in the comprehensive review of Edwards and Truelove or in the recent monograph on ulcerative colitis published by Goligher *et al* in 1968. The authors of this monograph, in discussing systemic complications, refer to the common mouth lesions found in ulcerative colitis, namely, aphthous ulceration and moniliasis. They record that 77

out of their 465 patients developed one or more mouth lesions at some stage of the illness. The vast majority of these cases were of aphthous ulceration and many of the patients recorded that the mouth ulcers would flare up together with their bowel symptoms. They recorded one case of aphthous ulceration and ulcerative pharyngitis associated with ulcerative colitis.

Our patient gave no history of aphthous disease in the past and the appearance of his mouth was entirely different from that of multiple aphthous ulceration.

The treatment of pyostomatitis vegetans is of interest. Several of the authors referred to in Table I recorded that the use of topical corticosteroids failed to give a permanent remission. Our patient, treated with oral prednisone, experienced a dramatic clearance of the mouth lesions. This response to systemic corticosteroid therapy suggests that the pyostomatitis vegetans, like the pyoderma gangrenosum, erythema nodosum, and uveitis associated with ulcerative colitis, is a disease process of the hypersensitivity type which readily responds to corticosteroids but which may equally readily break out again with or without an exacerbation of the colitis.

References

Edwards, F., and Truelove, S. C. (1964). The course and prognosis of ulcerative colitis. Pts. III & IV. *Gut*, 5, 1-22.

Evans, J. G., and Acheson, E. D., (1965). An epidemiological study of ulcerative colitis and regional enteritis in the Oxford area. *Gut*, 6, 311-324.

Goligher, J. C., DeDombal, F. T., Watts, J. McK., and Watkinson, G. (1968). *Ulcerative Colitis*, pp. 136-138. Bailliere London.

Hays, M. A. (1961). Pyostomatitis vegetans. Report of a case. *Oral Surgery*, 14, 1053-1056.

McCarthy, F. P. (1949). Pyostomatitis vegetans. Report of 3 cases. *Arch. Derm. Syph. (Chic.)*, 60, 750-764.

McCarthy, P., and Shklar, G. (1963). A syndrome of pyostomatitis vegetans and ulcerative colitis. *Arch. Derm.*, 88, 913-919.

Margoles, J. S., and Wenger, J. (1961). Stomal ulceration associated with pyoderma gangrenosum and chronic ulcerative colitis. *Gastroenterology*, 41, 594-598.

Zegarelli, E. V., and Kutscher, A. H. (1962). Oral 'Pyoderma'. Report of 2 cases. *Amer. J. dig. Dis.*, 7, 281-288.