CHRONIC ULCERATIVE COLITIS WITH SYSTEMIC LUPUS ERYTHEMATOSUS

Report of a Case

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THE occurrence of both chronic ulcerative colitis and systemic lupus erythematosus in the same patient has not to our knowledge been previously reported in the literature. At about the time we saw a patient in Cleveland with both diseases, Boone and McKee¹ in Toronto reported to us that they had observed and treated a similar case.

There is some evidence that chronic ulcerative colitis may be one of the collagen diseases, similar to lupus erythematosus. Levine, Kirsner, and Klotz² noted an absence of the homogeneous ground-glass substance of the basement membrane of the epithelial cells of the rectal mucosa in patients with chronic ulcerative colitis. They report: "The epithelial cells of the mucosa were morphologically intact . . . , but in many areas the mucosa had separated from the underlying connective tissue. In the intervening space a homogeneous, Hotchkiss-positive, metachromatic material was observed. Such areas were often free of inflammatory response." This metachromatic material was not present in a patient with active amebiasis and active inflammation or in a patient with lymphopathia venereum and active colitis. Finally, they found that biopsies in patients with ulcerative colitis who had received ACTH therapy showed areas in which the ground-glass substance of the basement membrane had returned. Warren and Sommers³ have described a vasculitis resembling periarteritis nodosa or thromboangiitis obliterans in some patients with chronic ulcerative colitis. These two studies^{2,8} suggest that chronic ulcerative colitis may be a systemetic, connective tissue or collagen disease similar to systemic lupus erythematosus, rather than a local infection of the colonic mucosa.

The occurrence of erythema nodosum, rheumatoid arthritis, iritis, and glomerulitis in association with ulcerative colitis suggests the aforementioned possibility of an etiologic relationship. Frequently these systemic complications

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become worse with an acute flare-up of the colitis, and subside as the colitis improves. Because of the similarity in some respects between chronic ulcerative colitis and systemic lupus erythematosus, and because of the apparently infrequent coexistence of the two conditions, we believe that this report of our patient having both diseases will be of interest.

Case Report

A 16-year-old girl was first examined on May 2, 1955; she had had frequent watery stools and abdominal cramps for the previous seven years, and bloody stools during the previous four years. In 1952, a barium enema examination, elsewhere, led to a diagnosis of ulcerative colitis. She had had severe anemia requiring four blood transfusions in 1951 and again in 1954. Episodes of fever occurred intermittently with a flare-up of colitis and arthritis. In the summer of 1954, she developed migratory joint pain, swelling, and joint tenderness for which she was given hydrocortisone, 40 milligrams daily. She continued to have diarrhea with three to ten loose stools daily. Occasionally, blood was present in the stools.

On physical examination the patient appeared to be malnourished, weighing only 92 pounds. The only significant findings were the fusiform swellings of the proximal joints of the hands, tenderness of the left knee, and numerous subcutaneous nodules typical of those found in patients having rheumatoid arthritis. There were apical and basal systolic murmurs (grade II) and some cardiac enlargement consistent with mitral insufficiency.

Pertinent laboratory findings were: hemoglobin, 12.1 gm. per hundred milliliters; white blood cells, 9300 per cu. mm. with 20 per cent eosinophils; sedimentation rate 1.75 mm. per minute; bromsulfalein dye, 14 per cent retention in 45 minutes; Wassermann and Kahn test results, negative; cephalin-cholesterol flocculation test, 4 plus; thymol turbidity, 20 units; urea clearance rate, 56 per cent in the first hour and 73 per cent in the second hour; blood urea, 24 mg. per hundred milliliters. All stools were negative for parasites. The agglutination test for dysentery organisms was negative; stool culture was negative for pathologic organisms. The Tiselius electrophoretic protein pattern was: albumin 2.66, alpha globulin 0.61, beta globulin 1.49, and gamma globulin 2.76 gm. per hundred milliliters. The tests for lupus erythematosus (L. E.) (peripheral blood, plasma-dog⁵) all were positive.

A proctoscopic examination disclosed a thickened and granular mucosa with fine ulcerations consistent with chronic ulcerative colitis and a fissure in ano. Barium enema examination showed ulcerative colitis that involved the entire colon and terminal ileum. A roentgenogram of the chest showed cardiac enlargement (left ventricular) and minimal bilateral pleural effusion.

The patient was treated with diet, hydrocortisone (20 mg. twice daily), chloroquine, Azulfidine,* Pyrodoxine, and isoniazid. On May 20 she was passing four stools a day but continued to have pain in the joints. On July 23 the arthritis had improved but she stated that she was unable to tolerate the Azulfidine.

She was seen again August 8 at which time the presenting complaint was a sudden pain in the right lower quadrant of the abdomen, nausea and vomiting, and increase in the diarrhea. Her temperature was 102 degrees F. There was a tender mass in the right lower quadrant. She was admitted to the hospital and treated with prednisone, 10 mg. three times daily, and with penicillin, 600,000 units daily. She improved daily; there was less diarrhea and the mass diminished in size. She was discharged from the hospital on

^{*}Azulfidine, Pharmacia Pharmaceutical Co., 270 Park Ave., New York, N.Y.

August 16, eight days after admission, and was advised to take prednisone, 5 mg. three times daily, and penicillin, 600,000 units daily.

When seen on August 23 she was making satisfactory progress. By September 21 she daily was having two to four stools without blood. She continued to take prednisone and she was allowed to go to school on a half-time schedule. On October 3 she was having only two stools daily; hemoglobin was 12.4 gm. per hundred milliliters and the L.E. test was negative; her weight was 98 pounds, and she was attending school on a full-time schedule without difficulty.

Discussion

Our patient had systemic disease with rheumatoid arthritis, cardiac involvement (mitral insufficiency), lupus erythematosus, and chronic ulcerative dolitis. The diverse and distinct signs and symptoms, namely those of arthritis, lupus erythematosus, and colitis, responded to steroidal therapy and supplemental measures; with these the arthritis cleared, the L. E. test became negative, and the diarrhea subsided.

Boone and McKee's¹ patient had ulcerative colitis, lupus erythematosus, and evidence of hepatic dysfunction and portal cirrhosis with markedly enlarged liver and spleen. In our patient the positive cephalin-cholesterol floculation and thymol turbidity tests may reflect changes in the serum proteins that are due to the lupus erythematosus rather than to hepatic dysfunction, although the increased bromsulfalein retention suggests impaired hepatic function. The impaired hepatic function may be secondary to the ulcerative colitis, resulting in chronic diarrhea, negative nitrogen balance and the loss of large amounts of protein in the stool.

The similarities in the two conditions already discussed and the association of both diseases in one patient might suggest a common etiologic factor. However, if there were a common etiologic factor in both conditions, one would expect both diseases to occur frequently in association. To see whether we were overlooking latent lupus erythematosus, tests for lupus erythematosus were performed in 20 other patients with chronic ulcerative colitis. These were negative. Since there is no published report heretofore, and we know of only the one other patient having both conditions, the incidence of the coexistence of chronic ulcerative colitis and systemic lupus erythematosus seems to be no greater than that which would be expected by chance.

Although ulcerative colitis and systemic lupus erythematosus rarely appear together, further studies concerning their possible relationship appear indicated. We believe that L. E. tests should be done in all patients having ulcerative colitis who exhibit any systemic complications, such as arthritis, iritis, or erythema nodosum.

Summary

Ulcerative colitis and systemic lupus erythematosus have much in common. The similarity in regard to the absence of the homogeneous ground-glass

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substance, the vasculitis in some patients and the systemic complications all suggest that ulcerative colitis as well as lupus erythematosus may be a systemic collagen disease.

The association of ulcerative colitis and lupus erythematosus is uncommon: we know of only two cases—our own and the one of Boone and McKee.¹ In our patient, chronic ulcerative colitis, lupus erythematosus, rheumatoid arthritis, and mitral insufficiency coexisted. The possibility of a common etiologic factor in ulcerative colitis and lupus erythematosus is only a matter of speculation at the present time.

References

- Boone, J., and McKee, J. A.: (Hospital for Sick Children, Toronto, Ontario, Canada): Personal communication, August 4, 1955.
- 2. Levine, M. D., Kirsner, J. B., and Klotz, A. P.: New concept of pathogenesis of ulcerative colitis. Science 114: 552-553 (Nov. 23) 1951.
- 3. Warren, S., and Sommers, S. C.: Pathogenesis of ulcerative colitis. Am. J. Path. 25: 657-679 (July) 1949.
- 4. Gonyea, L. M., Kallsen, R. A., and Marlow, A. A.: Occurrence of "L.E." cell in clotted blood. J. Invest. Dermat. 15: 11-12 (July) 1950.
- 5. Haserick, J. R., and Bortz, D. W.: Normal bone marrow inclusion phenomena induced by lupus erythematosus plasma. J. Invest. Dermat. 13: 47-49 (Aug.) 1949.