Case report

Therapeutic effect of intraarterial prednisolone injection in severe intestinal Behçet’s disease

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A 64-year-old woman with severe intestinal Behçet’s disease who was unresponsive to conventional therapies, including intensive intravenous steroid injections, underwent intraarterial steroid injection therapy. After the infusion of prednisolone into the mesenteric arteries, her colon ulcers improved markedly, and the frequency of bloody stools decreased immediately. The present case suggests that intraarterial steroid injection therapy may be potentially useful in severe intestinal Behçet’s disease.

Key words: Behçet’s disease, intraarterial injection therapy, prednisolone, steroid-induced pancreatitis

Introduction

Behçet’s disease is a multisystem disorder originally characterized by recurrent oral and genital ulcerations, ocular inflammation, and skin lesions. The disease also involves the articular, vascular, gastrointestinal, and nervous systems. Intestinal Behçet’s disease with gastrointestinal ulcers accounts for approximately 1%–2% of Behçet’s disease. This form of the disease is characterized by many punched-out ulcers in the intestine, and sometimes involves complications such as intestinal hemorrhage, perforation, and septic shock, the presence of which often leads to surgical resection of the lesion. While intraarterial steroid injection has been reported to be effective in patients with severe active ulcerative colitis, there has been no detailed report of this treatment in patients with intestinal Behçet’s disease. We successfully employed intraarterial steroid injection therapy and avoided surgical resection in a patient with intestinal Behçet’s disease. We report the case here and assess the potential utility of intraarterial steroid injection therapy in this disease.

A 64-year-old woman complaining of high fever and lumbago was referred to Gifu University Hospital. A diagnosis of Behçet’s disease was established, based on the presence of oral and genital ulcers, erythema nodosum, arthralgia, and ocular inflammation. The results of laboratory tests upon admission were as follows: hemoglobin (Hb), 6.5 g/dl; white blood cell count (WBC), 20 100/µl; and C-reactive protein (CRP), 15.3 mg/dl (normal, <0.4 mg/dl). The patient’s clinical course is shown in Fig. 1. Her general condition improved after treatment with 1 mg/day of colchicine, antibiotics, blood transfusion, and total parenteral nutrition. On the 11th hospital day, however, abdominal pain and bloody stools appeared suddenly. An upper gastrointestinal endoscopy showed multiple gastric ulcers and erosions (Fig. 2a). There were no specific histological findings in the gastric lesions, and only a moderate degree of inflammatory cell infiltration was observed in the gastric mucosa. Colonoscopy examination disclosed multiple punched-out ulcers between the rectum and transverse colon (Fig. 3a). Pathological examination of the endoscopic biopsy specimen showed nonspecific acute inflammation in the colon. The patient’s small intestine showed no remarkable findings in a double-contrast study. One gram/day of intravenous methylprednisolone, given for 3 days, and subsequent daily 40-mg doses of intravenous prednisolone improved the gastric lesions (Fig. 2b), but did not suppress the intestinal hemorrhage, and the patient’s Hb fell to 4.4 g/dl. Because intravenous injections of prednisolone sometimes cause delayed clinical effects in ulcerative colitis patients, we continued to wait until there was much intestinal hemorrhage. On confirming
the limitations of intravenous administration of prednisolone, intraarterial steroid injection therapy was chosen, prior to the intended resection of the lesion. After obtaining written informed consent from the patient, a 20-mg injection of prednisolone was delivered via the transfemoral route, using a standard catheter placed selectively into the superior and inferior mesenteric arteries, respectively. Although the patient's angiographic findings were almost normal, a slight capillary brush appeared (Fig. 4a,b). The external diameter of the inferior mesenteric artery was 4.5 mm.

Surface markers of lymphocytes were measured with a flowcytometer. Colchicine treatment has strong systemic effects, and affects lymphocyte subsets. The CD4-positive cell/CD8-positive cell ratios before and after the colchicine treatment were 0.89 and 4.4, respectively. The percentages of CD4-positive cells before and after the intraarterial prednisolone injection were 53.6% and 17.7%, respectively. The percentages of CD8-positive cells before and after this treatment were 12.2% and 3.2%, respectively. The CD4-positive cell/CD8-positive cell ratios before and after this treatment were 4.4 and 5.5, respectively. The peripheral white blood cell counts before and after the intraarterial injection therapy were 29 000/μl and 11 200/μl, respectively. The total lymphocyte count did not differ significantly before (2150/μl) and after (2300/μl) the intraarterial injection therapy.

On the day after the intraarterial prednisolone injection, the frequency of bloody stools decreased. WBC and CRP gradually returned to the normal ranges. However, discomfort in the epigastrium and the transient elevation of serum amylase (458 IU/l; normal, 73–230 IU/l), lipase (266 IU/l; normal, 7–48 IU/l), trypsin (1200 ng/ml; normal, 110–460 ng/ml), and phospholipase A2 (2620 ng/dl; normal, 130–400 ng/dl) were observed after the intraarterial steroid injection. Abdominal ultrasonography of the pancreas showed a homogeneous and moderate enlargement of the head of the pancreas, without any abnormality of vessels, bile, and pancreatic ducts, and no lymphadenopathy or peritoneal fluid collection. Colonoscopy was performed again, 12 days after the intraarterial steroid injection, and showed multiple healing ulcers, but no open ulcer between the rectum and transverse colon (Fig. 3b). The disease was considered to be in remission. The daily intravenous prednisolone was then tapered, in combination with an increasing dose of salazosulfapyridine, and this was finally replaced by 4 g/day of oral salazosulfapyridine.