Pneumatosis Cystoides Intestinalis in a Patient with Ulcerative Colitis

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A 72-year-old man developed ulcerative colitis (UC) at the age of 48 and remained in remission only by continuing salazosulfapyridine. Colonoscopy, performed at the age of 68, indicated that the patient was in remission; however, numerous hemispherical protuberant lesions with smooth surfaces extending from the sigmoid colon to the cecum were observed (Fig. 1). In addition, computed tomography (CT) showed numerous cysts in the low-absorption regions of the same segment of the colon (Fig. 2). Microscopic histological examination of a specimen from the ascending colon showed pneumatic cysts in the submucosa, architectural glandular disarray with atrophy, and mononuclear inflammatory infiltrate in the lamina propria (Fig. 3). The patient was subsequently diagnosed with pneumatosis cystoides intestinalis (PCI) in the setting of UC. As the patient was asymptomatic, he was followed up without additional treatment for PCI and colonoscopy was performed annually over the following 4 years. However, the PCI findings did not change. Because the patient was in remission, salazosulfapyridine was discontinued at the age of 72. Subsequently, the cysts in the intestinal wall disappeared from the CT images 3 months later, and a colonoscopy performed 5 months after therapy discontinuation evidenced almost complete disappearance of the hemispherical protuberant lesions.

Pneumatosis cystoides intestinalis is a rare disease characterized by numerous air-permeable cysts forming in the submucosal and serosal layers of the intestinal wall [1]. The most frequent causes are elevated intraluminal pressure, mucosal disruption, and increased mucosal permeability associated with drugs [2, 3]. There have been very few reports of cases of UC complicated by PCI [4-6], and steroids and immunosuppressive drugs are believed to be risk factors associated with PCI. However, in this case, since the PCI disappeared shortly following discontinuation of salazosulfapyridine, it can be inferred that the PCI was caused by salazosulfapyridine.

This is the first report of PCI following salazosulfapyridine use. Discontinuing salazosulfapyridine may be a treatment strategy in patients with UC who develop PCI.

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REFERENCES


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