

Fig. 1. Abdomino-pelvic CT finding. (A) It showed soft tissue infiltrations (arrow) surrounding the presacral space. (B) Associated hydronephroureterosis was noted.

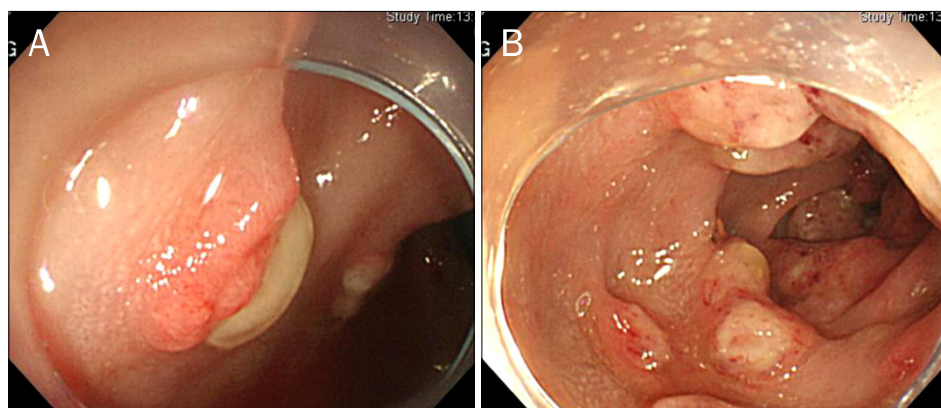


Fig. 2. Colonoscopic image. Sessile polyps with caps of purulent exudates were observed.

CASE REPORT

A 45-year-old woman with hematochezia and pain in both flanks was transferred to our hospital. She had experienced chronic constipation and straining on defecation for several years, which was occasionally managed with aloe and sitz baths. In September 2008, she had been admitted to another hospital for the evaluation of constipation, where colonoscopy revealed multiple polyps of various sizes with exudates from distal the rectum to the sigmoid colon. Microscopic examination of biopsy specimens from a large polyp showed serrated adenoma. Low anterior resection and loop ileostomy were performed due to the size of the polyp. Microscopic examination of the surgical specimens also showed serrated adenoma with low grade dysplasia: 6×4.5 cm in size, with involvement of the distal resection margin and regional lymph node of reactive hyperplasia without tumor cells. The patient's constipation was resolved after surgery. In December 2008, she underwent ileostomy takedown. After surgery, she

complained of lower abdominal pain and persistent mucoid diarrhea that occurred more than 10 times per day. After a few weeks of medical treatment, she was transferred to our hospital.

The patient's concentrations of acute-phase reactants were high: ESR, 64 mm/hour (normal: 0-27 mm/hour); CRP, 11.3 mg/dL (0-0.3 mg/dL). Enhanced abdomino-pelvic computed tomographic scans located ill-defined soft-tissue attenuations around the aortic bifurcation and presacral space, resulting in bilateral hydronephroureterosis (Fig. 1). On retrograde pyelography, both ureters were medially displaced and narrowed. Bilateral double-J stent insertion was performed to resolve both hydronephroses. Colonoscopy revealed multiple, variable reddish sessile polyps with white fibrinopurulent exudates (Fig. 2). Histopathologic evaluation revealed hyperplastic glands without dysplasia and exudates of the mucosal surface (Fig. 3).

The patient underwent laparoscopic retroperitoneal biopsy and adhesiolysis to exclude malignancy, resulting in a dif-

ferential diagnosis of retroperitoneal fibrosis. Microscopic examinations of surgical specimens revealed fibroblasts and

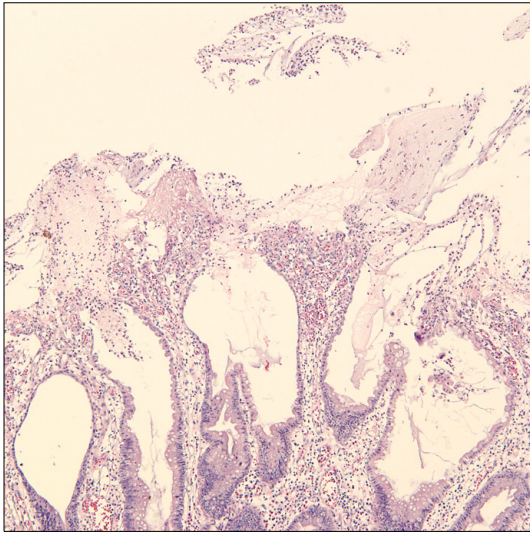


Fig. 3. Pathologic finding of colonoscopic biopsy. It showed the surface of polyps covered by exudates (H&E, ×100).

lymphocyte infiltration. We observed hyperplasia of numerous blood vessels and peripheral nerves due to secondary post-inflammatory changes (Fig. 4). Autoimmune disease and several other causes of secondary retroperitoneal fibrosis were excluded by blood tests. Based on these findings, we made a final diagnosis of cap polyposis with idiopathic retroperitoneal fibrosis.

The patient was treated with oral metronidazole for 10 days for *Helicobacter pylori* (*H. pylori*) eradication. However, her symptoms did not improve. An initial daily dose of prednisolone 50 mg was administered to treat the patient's retroperitoneal fibrosis. Ill-defined soft-tissue attenuation around the aortic bifurcation and presacral space were decreased at a follow-up enhanced CT examination 8 weeks after the initiation of treatment. Surprisingly, the patient's cap polyposis was much improved at follow-up colonoscopy 6 weeks after the treatment (Fig. 5).

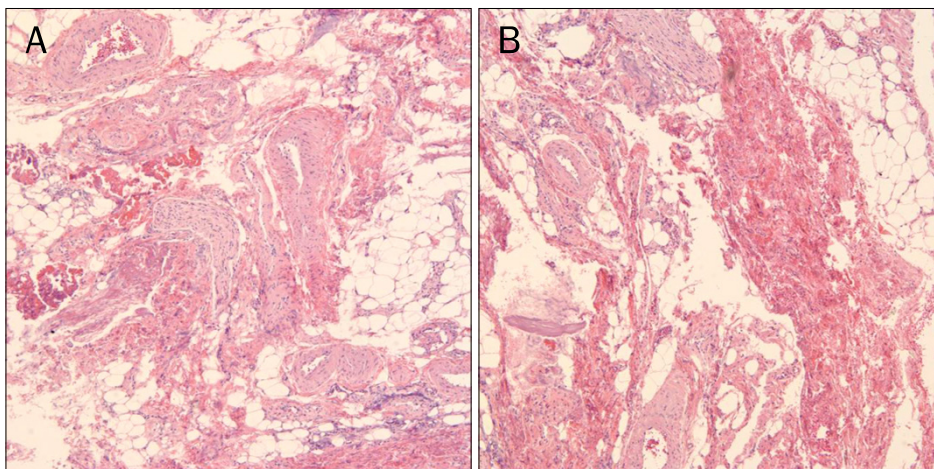


Fig. 4. Pathologic findings of retroperitoneal biopsy. (A) It showed fibroblast and lymphocyte infiltration in fibrosis (H&E, ×100). (B) Numerous blood vessels and peripheral nerves were observed due to secondary post-inflammatory change (H&E, ×100).

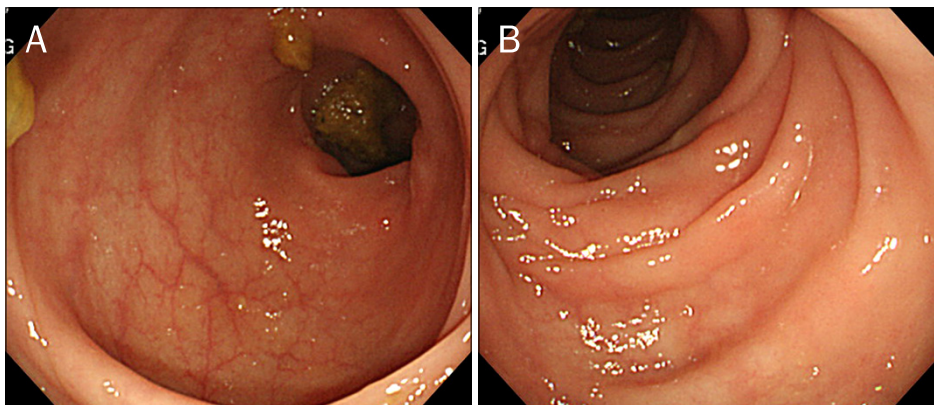


Fig. 5. Follow-up endoscopic images. It showed resolved cap polyposis.

DISCUSSION

Cap polyposis, which was first described in 15 patients by Williams et al.⁹ in 1985, is a benign colorectal condition characterized by multiple polyps covered with thick layer of fibrinopurulent exudates.

The pathogenesis is unclear. Local ischemia to the colonic mucosa due to abnormal colonic motility and repeated trauma caused by straining during defecation are important factors in the development of cap polyposis.¹⁰ Inflammatory bowel diseases and infectious agents such as *H. pylori* or *Escherichia coli* O18 are also possible causes in the absence of abnormal colonic motility.^{2,11} Similar to the present case, Cap polyposis has been described following pelvic surgery.²

The diagnosis of cap polyposis is established during colonoscopy by recognizing the typical colonoscopic findings of erythematous polyps with adherent mucoid caps.

Drug therapies to treat cap polyposis are usually unsuccessful. In our patient, treatment with a soluble fiber supplement and avoidance of straining during defecation were ineffective. Symptomatic improvement after treatment with metronidazole¹² alone or in combination with betamethasone enemas¹³ have previously been reported. Symptoms and polyps may also disappear after eradication therapy for *H. pylori*.¹⁴ However, optimal treatment had not been established. Interestingly, Ohkawara et al.⁴ reported spontaneous resolution of cap polyposis. Administration of budesonide enema was ineffective in our patient, and while *H. pylori* was identified in stomach biopsy specimens, eradication therapy was also ineffective. Patients with solitary cap polyps respond well to endoscopic polypectomy, while patients with multiple polyps and concurrent anorectal pathology require surgical resection.¹⁵ A previous study reported that four patients who underwent anterior resection were all symptom-free at a median of 48 months (range, 18-72 months) after surgery.¹⁵ However, cap polyposis may occur after pelvic surgery,² and symptomatic polyposis can also recur.^{3,16} Kim et al.¹⁷ reported a case of cap polyposis that was remarkably improved after a single infliximab infusion and did not recur for 3 years. Our patient underwent low anterior resection, and then developed symptomatic polyposis with bilateral ureteral obstruction due to retroperitoneal fibrosis.

Corticosteroids are the most useful drugs for the treatment of idiopathic retroperitoneal fibrosis. However, the opti-

mal dose and duration of steroid therapy are not established. In our patient, an initial daily dose of prednisolone 50 mg was administered.¹⁸

This is the first report of cap polyposis with retroperitoneal fibrosis after surgery. Our patient's cap polyposis was much improved after 6 weeks. The present case of cap polyposis was benign and had a self-limiting course.

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