Capsule-endoscopic findings in immunoproliferative small-intestinal disease

Capsule endoscopy is a noninvasive diagnostic method used in various gastro-intestinal diseases such as Crohn's disease and celiac disease. Immunoproliferative small-intestinal disease (IPSID) is a proliferative disorder of IgA-producing B lymphocytes [1]. IPSID has been characterized by the loss of the mucosal circular folds, a thickened, cobblestone appearance of the mucosa, and multiple sessile polyps on endoscopic examination [2]. As the disease progresses, tumors appear, usually in the proximal small intestine [3]. To the best of our knowledge, this is the first report of the findings from capsule endoscopy in IPSID.

A 38-year-old woman was admitted with vomiting, chronic diarrhea, and weight loss. Her medical history revealed that she had iron deficiency anemia and celiac disease; physical examination was recorded as normal except for local edema of her foot. Her body mass index (BMI) was 16 kg/m². Tests for IgA class endomysial antibody, IgA tissue transglutaminase antibody, and IgA and IgG antigliadin antibodies were negative. Circular aphthous ulcers were found on mucosal folds in the second part of the duodenum during an endoscopic examination. Map-shaped areas and mucosal depressions were found in the terminal ileum at colonoscopy. Histopathologic examination revealed focal villous atrophy in duodenal biopsies and villous atrophy and a diffuse, atypical lymphoid-cell infiltration in ileal biopsies. Immunohistochemical staining of the biopsy specimens with CD79, MUM-1, CD20, CD3, and MPO was positive, consistent with the diagnosis of IPSID. Abdominal computed tomography (CT) and CT-enterography did not reveal any abnormality.

The appearances at capsule endoscopy were loss of villi in the intestinal mucosa; small ulcers (● Fig. 1 a) and erosions in the proximal small intestine; fissuring, scalloping, and nodularity (● Fig. 1 b), mosaic pattern (● Fig. 1 c), layering

Fig. 1 Images from a capsule endoscopy in a woman with immunoproliferative small-intestinal disease (IPSID) showing: **a** an ulcer in the proximal small intestine; **b** fissuring and scalloping of folds and nodularity of the intestinal mucosa; **c** a mosaic pattern in the small intestine; **d** layering of the mucosal folds in the distal small intestine.

(● Fig. 1 d), and a few polypoid lesions in the distal small intestine.

The capsule-endoscopic findings of our patient were similar to the capsule-endoscopic findings in patients with celiac disease; however, in our patient serologic antibody tests for celiac disease were negative and she had no response to a gluten-free diet.

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Competing interests: None

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