A first report of collagenous gastritis, sprue, and colitis in a 9-month-old infant: 14 years of clinical, endoscopic, and histologic follow-up

A 9-month-old boy was admitted to hospital with acute severe dehydration because of watery diarrhea. He had a history of low weight for height, due to chronic intermittent vomiting and diarrhea since 15 days from birth.

Hypoproteinemia (3.5 g/100 ml), hypoalbuminemia (1.4 g/100 ml), hypogammaglobulinemia (4.6 g/l) and lymphocytosis (8.4 G/l) were found. Endoscopy showed an atrophic gastric mucosa with erosions, without other alterations.

Histology showed a diffuse irregular thickening of the subepithelial collagen tissue (SCT) (stomach 50 μ m, duodenum 12 μ m, colon 12 – 17 μ m), erosive epithelium, and jejunal villous atrophy (grade III).

In spite of 13 years of aggressive treatment (prednisolone $0.5-1 \, \text{mg/kg/d}$, alternating with budesonide $3-4 \, \text{mg/d}$, night enteral nutrition, and a gluten-free diet), clinicopathological disease progression was observed (**Pigs. 1, 2**).

Endoscopy at age 3 showed pseudopolyps with inflammatory areas in the gastric body, and erythematous mucosal areas in the colon. At age 10, a thick small tubular stomach, disappearance of gastric folds, and diffuse atrophic areas (as far as the duodenum), with erythema and multiple small submucosal nodules were observed. The colonic mucosa had become diffusely pale, thickened, and nodular, with disappearance of the normal vascular pattern. Histological examination showed a diffuse atrophic mucosa and an increase in the SCT (12-100 µm) was observed in the gastrointestinal tract. At age 14, because there was no clinicopathological improvement, the patient began total parenteral nutrition; this led to discontinuation of corticotherapy 2 months later and complete clinical improvement. Collagenous gastritis, sprue, and colitis are of unknown pathogenesis and are frequently associated with other disorders [1]. Diagnosis is on the basis of a strip SCT greater than 10 µm, with an irregular or focal distribution [1,2]. Symptoms vary according to the gastrointestinal segment affected [1-5]. Collagenous gastritis and colitis are rarely encountered in pediat-

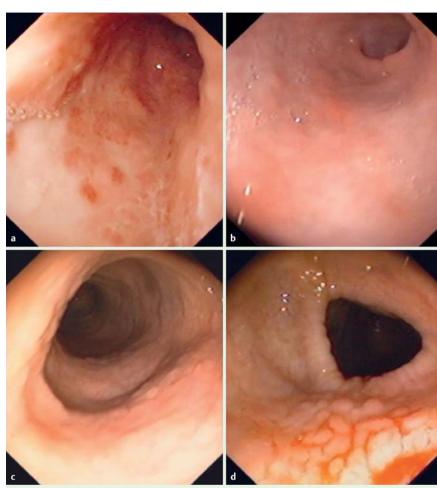


Fig. 1 Endoscopic appearance in 10-year-old boy with collagenous gastritis, sprue, and colitis: **a** qastric mucosa; **b** duodenal mucosa; **c** right colon; **d** transverse colon.

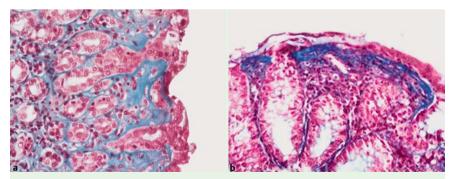


Fig. 2 Histological appearances: **a** gastric, showing the antral mucosa with lymphocytic infiltrate and collagen deposits; **b** colonic, showing the collagenous deposits (**a** and **b**, Masson's trichrome stain × 400.)

rics. Two cases have been reported where these conditions were associated [4,5], but there have been no previous reports where both entities were associated with collagenous sprue. As the physiopathology remains unknown, specific treatment is not available. The long-term outcome is still poorly documented [1]. This case demonstrates the complexity of the clinicopathological course caused by a diffuse gastrointestinal inflammation caused by collagenous deposition.

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