57-year-old male with a history of chronic alcoholism consulted our university medical center because of intermittent dysphagia for two years. Endoscopy, performed in another hospital, had not revealed any diagnosis. The patient occasionally suffered from unintentional stool loss. Proctological examination had shown no pathological findings, however neurologic examination had revealed a sensory neuropathy.

Barium-contrast esophagogram (Barium®, Guerbet, Sulzbach, Germany) showed multiple flask-shaped outpouchings in the esophageal wall. On double contrast esophagogram, most of the outpouchings were still filled with barium (Figure 1 and 2). A moderate stricture found in the upper esophagus (Figure 1) regressed slightly after the administration of butylscopolamin (Figure 2). Esophagogram depicted small interconnecting intramural tracks (Figure 1).

Endoscopic examination revealed multiple small diverticula in the esophageal wall (Figure 3). Histology revealed hyperplasia of the esophageal epithelia with signs of a moderate chronic esophagitis. Microbiological examination showed no pathological findings. Manometric examination revealed esophageal hypermotility. The patient received butylscopolamin for a symptomatic therapy.

Discussion

Esophageal intramural pseudodiverticulosis (EIP) is a rare disorder that is characterized on esophagogram by multiple flask-shaped outpouchings in the esophageal wall. The pseudodiverticula represent dilated excretory ducts of deep esophageal mucous glands.

Levine et al. reported an incidence of 0.15% of EIP on 14350 studied barium esophagograms.\textsuperscript{[1]} However, only about 200 cases of EIP have been published so far. Clinical presentation is in about 75% of the patients dysphagia.\textsuperscript{[3,4]} However, EIP
can also be an incidental finding on esophagogram.\textsuperscript{1,3,4}

The etiology of EIP remains unknown, however current data suggests that pseudodiverticulosis is not a primary disease of the esophagus, it is much more a consequence of chronic irritation by different causes like esophageal reflux, diabetes or chronic alcoholism.\textsuperscript{1,3,4} Esophageal inflammation has been reported in up to 90\% of the patients with EIP.\textsuperscript{5} Dilatation of the ducts might be caused by obstruction with inflammatory material or extrinsic compression due to periductal inflammation with fibrosis.\textsuperscript{5} Disturbance of neurologic functions, e.g. diabetes mellitus can cause esophageal hypermotility, an entity that has only been reported in three cases of EIP.\textsuperscript{3,4} Additional chronic neural involvement, as in our patient, has not been described before. We consider the impaired neurologic function as causative factor, however, we do not have an evidence for a direct cause-effect relationship.

Patients often have associated strictures, mostly in the upper esophagus. Endoscopic bouginage of associated strictures is effective in treating dysphagia. Treatment of the underlying esophageal disease is mandatory, e.g., proton pump inhibitors in esophageal reflux disease, or (as in our case) butylscopolamin for motility disorders.\textsuperscript{1,4}

Intramural tracking is often seen in patients with EIP.\textsuperscript{5} Although there is little known clinical significance of this finding, it is important in establishing differential diagnosis as intramural tracks should not be mistaken for ulceration or extramural barium collections associated with perforation.\textsuperscript{5} The most important differential diagnosis is underlying esophageal carcinoma, which has to be excluded carefully.\textsuperscript{1,6} Plavsic \textit{et al.} reported a much higher incidence of EIP in patients with esophageal carcinoma (4.5\% of 245 patients) than in a control group (0.9\% in a population of 6400).\textsuperscript{5} This implies increased risk of esophageal carcinoma, although a causative relationship could not be established by the study.

\textbf{References}