Moderate anemia (hemoglobin 97 g/L, MCV 84 fL, ferritin 22.1 µg/L) was detected on a follow-up visit to rheumatology. We performed an esophagogastroduodenoscopy and colonoscopy in search of bleeding with physiological findings. In accordance with ESGE guidelines (6) VCE was scheduled, an additional push enteroscopy (as required before VCE by the insurance company in our region) with biopsy showed stable findings without villous atrophy (Marsh 1, $CD3^+$ $CD8^+$ as shown by immunohistochemistry). The VCE revealed exulcerated, stenotized infiltration of the small bowel, with signs of bleeding (Fig. 1a, 1b), in front of which the capsule lodged. The capsule was visualized on an abdominal x-ray (Fig. 2). No infiltration was evident on a simultaneously performed CT enterography, only the position of the lodged capsule indicated the site of stenosis (Fig. 3). The patient was operated upon with a finding of infiltration of the jejunum at the mesenterial site, with bilateral lymphadenopathy. A resection of 60 cm of the jejunum (Fig. 4) with side-to-side anastomosis was performed. The patient was discharged 7 days after surgery without any postoperative complications. The specimen was evaluated by a histopathologist as moderately differentiated adenocarcinoma (Grade 2), with angioinvasion and a positive lymph node (1/13), R0 resection, pT3N1M0 (Fig. 5). The patient received adjuvant chemotherapy XELOX (capecitabine plus oxaliplatin) due to risk factors (positive lymph node and angioinvasion).

Fig. 2. *A visible capsule on the abdominal x-ray*

Fig. 3. *CT enterography (coronal scan) – VCE lodged in the jejunum with no evident CT signs of tumor infiltration*

Discussion

Malignant tumors of the small bowel represent only 1–3% of gastrointestinal malignancies. Adenocarcinomas are the most frequent finding (35–50%) followed by carcinoid tumors (30%), lymphomas (15%) and gastrointestinal stromal tumors other than sarcomas (10%) (7–9). Nevertheless, patients with CD are at an increased risk of some of these malignancies. Studies investigating the risk of development of carcinoma in patients with CD bring very different results, ranging from a 10-fold increased risk (2) to an 82.6-fold increased risk (7). Although the relative risk of lymphoma dramatically decreases after one year of gluten-free diet (RR 157.6 to 12.7), the decrease in cases of carcinoma is not so expressed (RR 38.0 to 6.4) and the p-value for the trend doesn't reach statistical significance (10).

Diagnosis of CD at an advanced age, untreated CD and possibly also persisting villous atrophy and male gender were described as risk factors of malignant complication development in celiac patients (11–14), but the number of patients in studies is usually small and the results aren't uniform. Our patient was diagnosed with CD at the age of 53 years. Due to the lack of symptoms it is possible that CD was undiagnosed and untreated for a long time in our patient, highlighting the importance of family screening in such cases. A high turnover of the inflammatory population with mucosal lymphocyte infiltration, an increased permeability to oncogenic factors, a malabsorption of protective substances such as vitamins A and E, or an impaired immune surveillance are considered as pathogenetic mechanisms of cancerogenesis in these settings (14). Another question is the course of development of the carcinoma. A majority of opinions lean toward adenoma-carcinoma evolution sequence (15, 16), but there are some studies which question this approach, providing cases suggestive of dysplasia development in the flat mucosa (3). In contrast to small bowel adenocarcinoma in general, where the duodenum is affected most frequently, carcinomas tend to develop in the jejunum in most cases of CD patients, which corresponds with our case. Vomiting, anemia, weight loss, intestinal bleeding, abdominal mass, and perforation are the most frequent symptoms (17). Unfortunately, a diagnosis is usually made in the advanced stages of the disease (74% in stage III-IV) (14). In most cases the carcinoma isn't accessible to esophagogastroduodenoscopy. CT or MR enterography and PET/CT are used