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# **Ileal pouch adenocarcinoma after restorative proctocolectomy for familial adenomatous polyposis**

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A 51-year-old male patient with a familial adenomatous polyposis (FAP) and a history of two-staged restorative proctocolectomy (RPC) with a stapled ileal pouch-anal anastomosis (IPAA) performed fifteen years earlier was admitted to our department with a suspicion of malignancy within the anal transitional zone (ATZ). Histopathological examination of colon specimens harvested during the primary surgery excluded the presence of malignancy. The patient did not appear on the follow-up visits since the operation. The patient had suffered from anorectal pain and anal bleeding for six months before admission. Rectoscopy revealed an ATZ tumor which was identified as adenocarcinoma of G2 differentiation in histopathologic examination (Figure 1A). Magnetic resonance imaging (MRI) showed a tumor infiltration into the mesorectum, sphincter complex, prostate, and the base of the penis (Figure 1B). Computed tomography (CT) showed no lymphatic or distant metastases. The

extra-intestinal manifestation of FAP was excluded in imaging (CT and MRI). The patient was qualified for neoadjuvant radio- and chemotherapy. After treatment completion the control MRI demonstrated significant tumor regression and the patient was qualified for surgery. A total abdominoperineal excision (Miles procedure) with the ileal pouch and sphincter resection was successfully performed – radical R0 resection with negative histopathologic margins (Figure 1C-D). There was no indication for further adjuvant chemotherapy. The postoperative course was uneventful. A twelve-month follow-up imaging showed no malignancy reoccurrence within the tumor field. However, several liver metastases were detected. Finally, the patient was qualified for the FOLFOXIRI chemotherapy regimen (folinic acid, fluorouracil, oxaliplatin, and irinotecan). The patient did not respond effectively to applied chemotherapy. Due to the lack of available treatment options and impaired general condition, the anticancer therapy was discontinued. The patient died several weeks later.

RPC is the established method for FAP patients. Some controversy exists about the proper pouch-anal anastomosis technique due to the risk of ATZ malignancy development [1]. Pouch neoplasia is a rare long-term complication diagnosed in FAP patients undergoing RPC [2-3]. A few risk factors of ileal pouch neoplasia were identified: preservation of the residual rectal mucosa, a time interval of over ten years from RPC, and patients over 44 years of age [2,4]. These budding risk factors were present in this case. The reported risk of pouch adenoma development in FAP patients after RPC at 5, 10, and 15 years after the procedure was estimated as 7%, 35%, and 75%, respectively [5]. Long-term endoscopic surveillance in FAP patients after RPC may allow for early-stage diagnosis of pouch neoplasia. The diagnosis of pouch neoplasia is a clinical challenge due to the long asymptomatic course and mimicking by other entities including pouchitis, which is the most common pouch-related complication [4-5].

The risk of IPAA adenocarcinoma is very high even many years after the initial surgical procedure, therefore, lifelong follow-up and periodic pouch surveillance should be routinely performed.

## **Article information**

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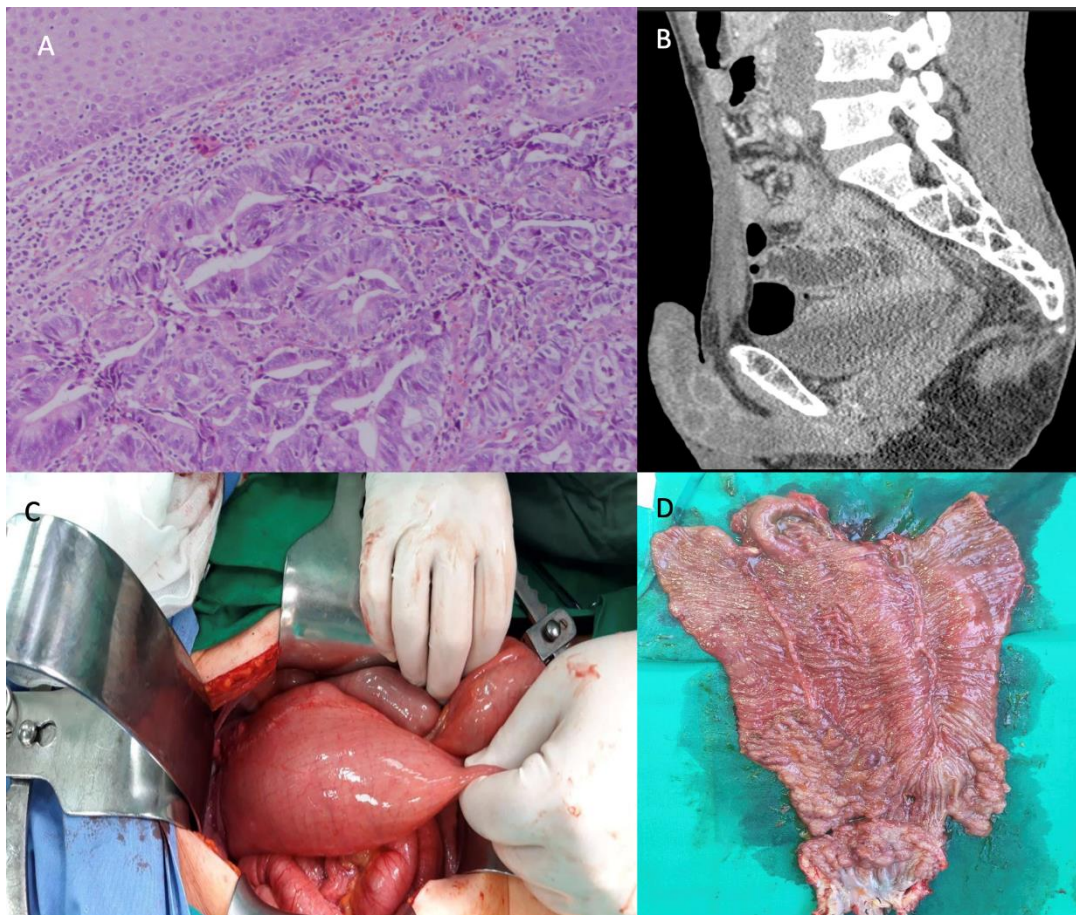
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**Figure 1** Familial adenomatous polyposis patient with advanced adenocarcinoma in ileal pouch 15 years after initial operation: **A** – microscopic image of adenocarcinoma G2 from ileal pouch taken during rectoscopy; **B** – magnetic resonance imaging of the abdomen and pelvis - tumor infiltrating the mesorectal tissue with invasion of the sphincter muscles, as well as the prostate and the base of the penis; **C, D** – intraoperative image of the ileal pouch. During and after resection

**Short title:** Cancer in the anal transitional zone after restorative proctocolectomy