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Low-Grade Appendiceal Mucinous Neoplasms: Description of Two Cases and Literature Review

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Aim: to present two clinical cases of low-grade appendiceal mucinous neoplasms (LAMN) and evaluate the oncological adequacy of ileocecectomy with ileoascending anastomosis in patients with cecal involvement.
Key points. Appendiceal mucinous neoplasms are rare tumors detected either during radiological examinations conducted for other reasons, intraoperatively in patients operated on with a diagnosis of appendicitis, or during histopathological examination of the appendectomy specimen. In 2025, two male patients, aged 36 and 70 years, with the aforementioned diagnosis were operated on at the Oncology Clinic of Azerbaijan Medical University. The neoplasm in the 36-year-old patient was discovered during intraoperative revision for suspected appendicitis at a district hospital. In the other patient, the tumor was detected preoperatively during a CT scan performed to stage an endoscopically and histopathologically confirmed sigmoid colon carcinoma. Both patients underwent an ileocecectomy with an ileoascending anastomosis for the appendiceal mucinous neoplasms. The postoperative period was uneventful, and histopathological examination of the resected specimens confirmed a low-grade appendiceal mucinous neoplasm (LAMN) in both cases.
Conclusion. In patients with LAMN adherent to or infiltrating the cecum, an ileocecectomy with ileoascending anastomosis can be performed as an adequate surgical procedure. Although this volume of surgery is less extensive than a right hemicolectomy, it can theoretically be compared with it in terms of providing oncological radicality.
Keywords: appendiceal mucinous neoplasms, low-grade appendiceal mucinous neoplasm, high-grade appendiceal mucinous neoplasm, mucocele, ileocecectomy
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Муцинозная опухоль червеобразного отростка низкой степени злокачественности: описание двух клинических случаев и обзор литературы

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Цель: представить два клинических случая муцинозных новообразований аппендикса низкой степени злокачественности (low-grade appendiceal mucinous neoplasms, LAMN) и оценить онкологическую адекватность илеоцекэктомии с илеоасцендоанастомозом у пациентов с вовлечением слепой кишки.
Основные положения. Муцинозные новообразования аппендикса — это редкие опухоли, которые выявляются либо в ходе радиологических исследований, проводимых по другим показаниям, либо интраоперационно у пациентов, оперируемых с диагнозом «аппендицит», либо при патогистологическом исследовании операционного материала после аппендэктомии. В 2025 г. в онкологической клинике Азербайджанского медицинского университета были прооперированы два пациента мужского пола в возрасте 36 и 70 лет с вышеуказанным диагнозом. Новообразование у 36-летнего пациента было обнаружено во время интраоперационной ревизии по поводу подозрения на аппендицит в районной больнице. У другого пациента опухоль была выявлена на предоперационном этапе при компьютерной томографии, выполненной с целью стадирования эндоскопически и гистологически верифицированной аденокарциномы сигмовидной кишки. Обоим пациентам по поводу данных муцинозных новообразований аппендикса была выполнена илеоцекэктомия с формированием илеоасцендоанастомоза. Послеоперационный период протекал без осложнений, а патогистологическое исследование резецированных макропрепаратов подтвердило диагноз LAMN в обоих случаях.
Заключение. У пациентов с LAMN, прилежащей к слепой кишке или инфильтрирующей ее, выполнение илеоцекэктомии и илеоасцендоанастомоза может рассматриваться как адекватное хирургическое вмешательство. Хотя данный объем операции менее обширен, чем правосторонняя гемиколэктомия, теоретически он сопоставим с ней с точки зрения степени радикальности онкологического лечения.

Ключевые слова: муцинозные опухоли червеобразного отростка, муцинозная опухоль низкой степени злокачественности, муцинозная опухоль высокой степени злокачественности, мукоцеле, илеоцекэктомия
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Introduction

Appendiceal mucinous neoplasms (AMNs) are a rare and heterogeneous group of neoplasms, with an age-adjusted incidence of approximately one to two cases per 1,000,000 population per year [1, 2]. Since these tumors are rare and lack typical clinical symptoms, they are usually discovered during radiological examinations performed for other indications or during intraoperative exploration in patients undergoing surgery for other pathologies. According to the 2016 consensus classification of the Peritoneal Surface Oncology Group International (PSOGI) Executive Committee and the 2019 World Health Organization (5th edition) system, AMNs are divided into 3 main categories (excluding serrated and hyperplastic polyps): low-grade appendiceal mucinous neoplasm (LAMN), high-grade appendiceal mucinous neoplasm (HAMN), and mucinous adenocarcinoma [1]. These categories can only be differentiated from each other (especially LAMN from HAMN) on the basis of histopathological examination, as the exact type of neoplasm cannot be determined based on radiological findings.

The aim of this study is to contribute to the literature on these tumors by presenting the outcomes of an ileocecectomy with an ileoascending anastomosis performed in two patients.

Case 1

A 70-year-old male patient was admitted to the Oncology Clinic of Azerbaijan Medical University with complaints of abdominal distension, constipation, and dull pain in the right iliac fossa. The patient was initially referred to another hospital with these complaints, and a colonoscopy performed there detected a tumor in the sigmoid colon. Histopathological examination of the biopsy specimens taken from the tumor confirmed a moderately differentiated adenocarcinoma. Contrast-enhanced abdominal CT performed for clinical staging showed no invasion of adjacent organs, no regional lymphadenopathy, and no suspicious metastatic foci in the liver. Additionally, the CT scan revealed an enlargement of the appendix up to 4.0 cm in diameter (Fig. 1).

The tumor was radiologically assessed as an AMN. Laboratory investigations revealed no abnormalities in vital organ functions. The serum CEA level was 2.86 ng/mL, and the CA 19-9 level was 12.6 U/mL. After discussion by a multidisciplinary team, the patient was scheduled for open surgery. During intraoperative exploration, a tumor was palpated in a 3.0–3.5 cm segment of the sigmoid colon with no visual evidence of serosal invasion. No suspicious metastatic foci were found on the peritoneal surface or in the liver upon visual inspection or palpation. The 12.0-cm-long appendix was enlarged to approximately 4.0 cm in diameter, had a carrot-like shape, was firm, and its tip projected towards the anterior abdominal wall (Fig. 2).

Although the tumor did not infiltrate the cecum, its base adhered directly to the cecal wall. Several loops of the terminal ileum were fixed to the appendix via dense adhesions. After careful sharp dissection of the fibrous bands, a sigmoid colonectomy with colocolic anastomosis was performed for the carcinoma, followed by an ileocecectomy with an ileoascending side-to-side anastomosis for the AMN.

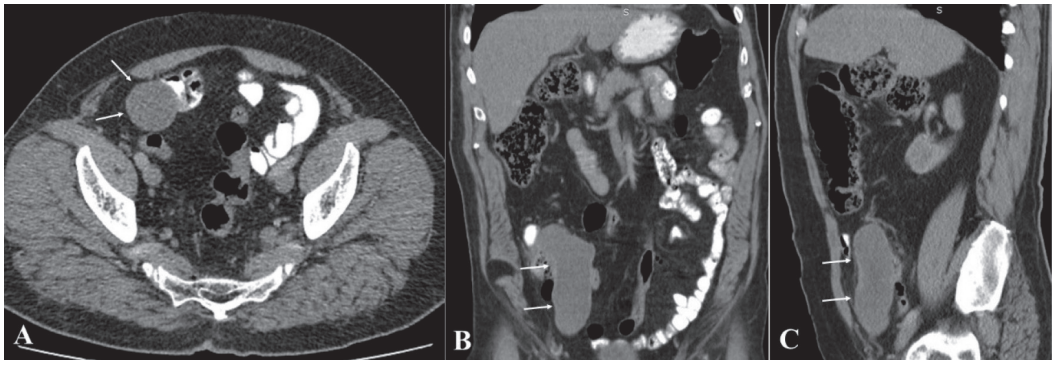

Figure 1. CT images of an AMN (arrows): A — axial view; B — coronal view; C — sagittal view

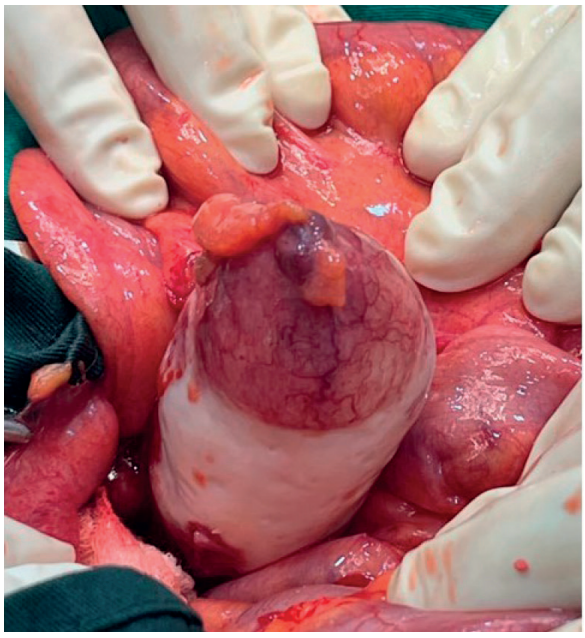

Figure 2. Visual appearance of an AMN

The ileocecectomy was performed by resecting the distal 15 cm of the ileum with its mesentery in a wedge-shaped fashion along the ileocolic artery. Thus, both procedures included dissection of the corresponding regional lymph nodes. The postoperative period was uneventful, and the patient was discharged in good condition on postoperative day 10. Histopathological examination of the resected specimens revealed a moderately differentiated adenocarcinoma of the sigmoid colon invading the subserosa, with no metastasis detected in the regional lymph nodes (pT3N0), and a concurrent low-grade appendiceal mucinous neoplasm (LAMN) (Fig. 3). The patient did not receive adjuvant chemotherapy. At the 6-month follow-up abdominal CT scan, no signs of recurrence or metastasis were detected.

Case 2

A 36-year-old male patient was referred to the Oncology Clinic of Azerbaijan Medical University from a regional hospital following a laparotomy with a diagnosis of an appendiceal tumor. The patient had been admitted to the regional hospital 12 days prior with a diagnosis of appendicitis. During intraoperative exploration, a mucocele originating from the appendix was discovered. The abdominal cavity was closed without any intervention on the tumor, and the patient was referred to a specialized clinic upon discharge. At our Oncology Clinic, the patient underwent a follow-up abdominal CT scan, which revealed an appendiceal neoplasm measuring approximately 2.0 cm in diameter and 3.0 cm in length, consistent with an AMN (Fig. 4).

The serum CEA level was 1.37 ng/mL, and the CA 19-9 level was 11.2 U/mL. Laboratory investigations revealed no abnormalities in vital organ functions. After discussion by a multidisciplinary team, the patient was scheduled for surgical resection. After laparotomy, loose postoperative adhesions in the ileocecal region were divided, revealing an appendiceal mucocele of the aforementioned size. The base of the mucocele was intimately attached to the cecal wall, and the capsule remained intact. No signs characteristic of tumor seeding or mucinous dissemination were detected during visual inspection and palpation of the peritoneal cavity. The patient underwent an ileocecectomy with a side-to-side ileoascending anastomosis. The ileocecectomy was performed by resecting the distal 15 cm of the ileum with its mesentery in a wedge-shaped fashion along the ileocolic artery. Thus, regional lymphadenectomy was also performed. The postoperative period was uneventful, and the

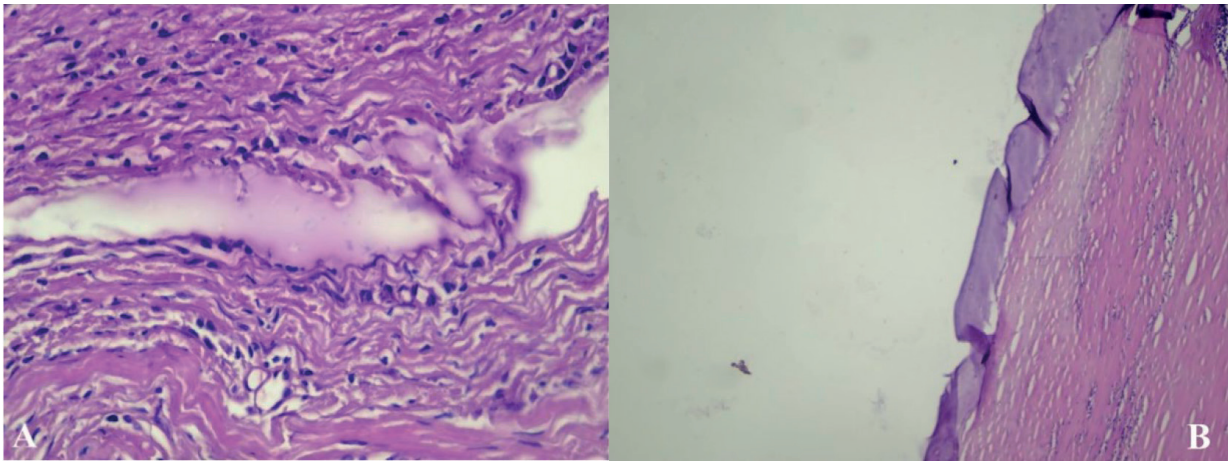

Figure 3. Microphotographs of the LAMN: A — acellular mucin in the muscularis propria (H&E, ×800); B — acellular mucin outside the appendix (H&E, ×200)

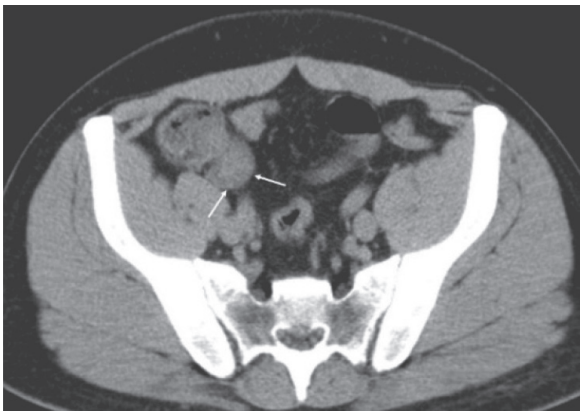

Figure 4. CT image of an AMN in the 36-year-old male patient (arrows)

patient was discharged in good condition on post-operative day 9. Histopathological examination of the resected specimen revealed a low-grade appendiceal mucinous neoplasm (LAMN) (Fig. 5). At the 3-month follow-up, no signs of recurrence or metastasis were detected.

Discussion

AMNs are rare, unique tumors found in approximately 0.8–1 % of patients undergoing appendectomy and accounting for 1 % of all colorectal malignancies [3–7]. More than 50 % of the tissue of these neoplasms is composed of extracellular mucin [8]. LAMN, a category of AMN, is a distinct oncological entity that frequently presents as a mucocele [4]. Specifically, an appendiceal mucocele is a clinical term that refers to the cystic dilatation of the appendix, which is most often caused by a LAMN. The literature reports that AMNs are more common in women than in men and are most frequently diagnosed in the sixth decade of life [3, 9]. Many patients present with symptoms consistent with acute appendicitis [9, 10]. Approximately half of the cases are asymptomatic at diagnosis and are discovered incidentally during examinations performed for other indications [11, 12]. Radiologically, an appendiceal diameter greater than 1.5 cm, wall thickening, or a soft tissue mass in the corresponding location suggests a mucinous neoplasm [9]. Noteably, there is no correlation between the diameter of the appendix and the histopathological grade of the tumor [4].

The 2019 World Health Organization (5th edition) classification system reinforces the 2016 PSOGI consensus classification, explicitly discouraging the use of confusing terminology such as mucinous cystadenoma of the appendix, mucinous tumor of uncertain malignant potential, borderline tumor of the appendix, mucinous cystadenocarcinoma of the appendix, replacing these names with the term “appendiceal mucinous neoplasm” [13].

Although the 2016 PSOGI consensus and the 2019 World Health Organization (5th edition) system present LAMN and HAMN as distinct entities separate from carcinoma, the UICC TNM classification categorizes AMNs that have not invaded the submucosa as Tis carcinoma. Mucinous neoplasms that have invaded the submucosa are classified as T1, and invasion into the muscularis propria, subserosa, serosa, and adjacent anatomical structures is classified as T2, T3, and T4, respectively, in accordance with the general rules of the TNM classification of gastrointestinal tract carcinomas. Although the T categories for HAMNs are determined according to the rules mentioned, the T1 and T2 categories for LAMNs are not distinguished at all, and the neoplasm is classified as Tis even if it invades the muscularis propria [14]. In other words, with respect to T categories, LAMN differs from other tumors of the gastrointestinal tract with that it does not have T1 and T2 categories.

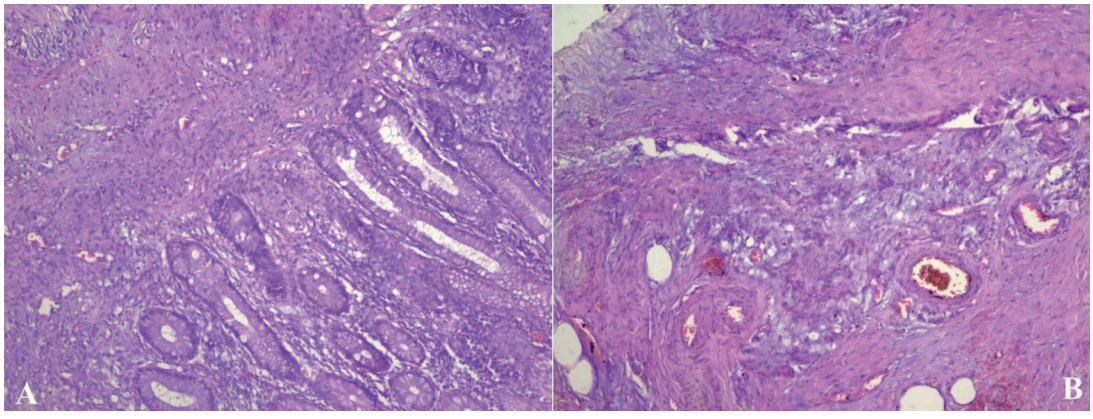

Рисунок 5. Microphotographs of the LAMN: A — submucosal fibrosis; B — acellular mucin in the muscularis propria (H&E, ×200)

The 2016 consensus by the PSOGI Executive Committee defined LAMN as a mucinous neoplasm with low-grade cellular atypia and at least one of the following features: loss of the muscularis mucosae, submucosal fibrosis, expansive (or diverticulum-like) growth through the appendiceal wall, intramural acellular mucin, undulating or flattened epithelial growth, rupture of the appendix, or mucin (acellular or cellular) outside the organ [15].

If left untreated, LAMN ruptures, potentially leading to pseudomyxoma peritonei. This complication is associated with a high mortality rate [16]. Typically, patients with AMNs seek medical attention when clinical signs resembling acute appendicitis appear or upon mucocele rupture. In cases where the neoplasm is confined to the appendix, preoperative diagnosis remains difficult [8]. Various surgical procedures are performed for the treatment of AMNs, including appendectomy, appendectomy with partial resection of the cecum, ileocecectomy, and right hemicolectomy [4]. Although some studies suggest that a right hemicolectomy does not demonstrate a superior survival rate comparing to an appendectomy in the treatment of AMNs, many centers consider an appendectomy for LAMN and a right hemicolectomy for HAMN to be the most adequate surgical procedures [8, 17]. However, as noted above, there are no radiological signs to differentiate between LAMN and HAMN, this distinction is made solely on the basis of histopathological examination. Therefore, determining the optimal extent of surgical intervention for AMNs detected either preoperatively or intraoperatively remains a challenging issue.

For appendiceal mucoceles, a laparoscopic approach is generally avoided due to the high risk of iatrogenic rupture. If the neoplasm is detected incidentally during laparoscopy, conversion to an open laparotomy is indicated. Laparotomy is also preferred because it allows for a careful inspection of other abdominal organs, especially the colon and ovaries, which are potential sites for concurrent primary mucinous tumors. In general, an appendiceal mucocele is associated with a mucinous tumor of the colon or ovary, occurring either synchronously or metachronously, in 11–20 % of cases [18]. Since the risk of colorectal cancer is high in patients with AMNs, a perioperative colonoscopy is strongly recommended [7, 15]. In one of our two patients, a synchronous sigmoid colon cancer was detected, and simultaneous surgery was performed for both tumors.

Since AMNs are rare and present with nonspecific symptoms, and clinicians frequently lack experience with them, these tumors are seldom considered in the differential diagnosis of patients with acute lower abdominal pain. These neoplasms are occasionally detected incidentally during radiological examination of the abdominal organs. When the condition is interpreted as acute appendicitis by a radiologist, the neoplasm is identified intraoperatively by the surgeon during an appendectomy. Understanding the nature of this condition, its histological types, clinical course, and prognosis is essential for making optimal therapeutic decisions in patients with an incidentally detected preoperative or intraoperative neoplasm. The description of such cases will contribute to improving clinicians' awareness of AMNs.

Since preoperative and intraoperative grading of AMNs is not feasible, ileocecectomy should be considered the procedure of choice for neoplasms intimately adherent to the cecum or extending to the cecal wall. This volume of surgery allows for a resection margin at a sufficient distance from the proximal border of the tumor. Compared to a right hemicolectomy, this procedure is less extensive while ensuring an R0 resection and the removal of regional lymph nodes along the ileocolic artery. Since the LAMN was in close contact with the cecal wall in both patients, appendectomy was not performed; instead, an ileocecectomy with an ileoascending anastomosis was executed. In the short-term postoperative period, neither patient experienced complications; furthermore, follow-up abdominal CT scans performed at 3 and 6 months, respectively, revealed no signs of local recurrence or metastasis.

Conclusion

In patients with LAMN adherent to or infiltrating the cecum, an ileocecectomy with ileoascending anastomosis can be performed as an adequate surgical procedure. Although this volume of surgery is less extensive than a right hemicolectomy, it can theoretically be compared with it in terms of providing oncological radicality.

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