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World J Clin Cases 2024 January 16; 12(2): 236-465



EDITORIAL

- 236 Use of artificial intelligence in the field of pain medicine
Chang MC

ORIGINAL ARTICLE**Retrospective Study**

- 240 Ultrasound blood flow characteristics changes in fetal umbilical artery thrombosis: A retrospective analysis
Hong SJ, Hong LW, He XQ, Zhong XH
- 249 Electroencephalogram findings in 10 patients with post-stroke epilepsy: A retrospective study
Wen LM, Li R, Wang YL, Kong QX, Xia M
- 256 Exploration of cardiac rehabilitation nursing for elderly patients with myocardial infarction based on individualized cardiac rehabilitation
Liu HN, Gao B
- 267 Survival benefit of concurrent chemoradiotherapy for advanced ampulla of Vater cancer
Kwon CH, Seo HI, Kim DU, Han SY, Kim S, Lee NK, Hong SB, Ahn JH, Park YM, Noh BG
- 276 Utility of plasma D-dimer for diagnosis of venous thromboembolism after hepatectomy
Miyake T, Yanagimoto H, Tsugawa D, Akita M, Asakura R, Arai K, Yoshida T, So S, Ishida J, Urade T, Nanno Y, Fukushima K, Gon H, Komatsu S, Asari S, Toyama H, Kido M, Ajiki T, Fukumoto T
- 285 Lenvatinib combined with sintilimab plus transarterial chemoembolization as first-line treatment for advanced hepatocellular carcinoma
Sun SS, Guo XD, Li WD, Chen JL

Observational Study

- 293 Timing theory integrated nursing combined behavior change integrated theory of nursing on primiparous influence
He YX, Lv Y, Lan TT, Deng F, Zhang YY
- 302 Inverse relationship between platelet Akt activity and hippocampal atrophy: A pilot case-control study in patients with diabetes mellitus
Tokuda H, Hori T, Mizutani D, Hioki T, Kojima K, Onuma T, Enomoto Y, Doi T, Matsushima-Nishiwaki R, Ogura S, Iida H, Iwama T, Sakurai T, Kozawa O

Randomized Controlled Trial

- 314 Impact of continuous care on cardiac function in patients with lung cancer complicated by coronary heart disease
Gao T, Luo JL, Guo P, Hu XW, Wei XY, Hu Y

- 322 Use of cognitive-behavioral career coaching to reduce work anxiety and depression in public employees
Otu MS, Sefotho MM

META-ANALYSIS

- 335 Efficacy and safety of Yangxue Qingnao Granules in treatment of migraine: A systematic review and meta-analysis
Zhou B, Wang GS, Yao YN, Hao T, Li HQ, Cao KG

CASE REPORT

- 346 Use of MLC901 in cerebral venous sinus thrombosis: Three case reports
Arsovska AA, Venketasubramanian N
- 354 Primary biliary cholangitis presenting with granulomatous lung disease misdiagnosed as lung cancer: A case report
Feng SL, Li JY, Dong CL
- 361 Asymptomatic low-grade appendiceal mucinous neoplasm: A case report
Yao MQ, Jiang YP, Wang YY, Mou YP, Fan JX
- 367 Surgically treating a rare and asymptomatic intraductal papillary neoplasm of the bile duct: A case report
Zhu SZ, Gao ZF, Liu XR, Wang XG, Chen F
- 374 Absence of enhancement in a lesion does not preclude primary central nervous system T-cell lymphoma: A case report
Kim CS, Choi CH, Yi KS, Kim Y, Lee J, Woo CG, Jeon YH
- 383 Mental retardation, seizures and language delay caused by new SETD1B mutations: Three case reports
Ding L, Wei LW, Li TS, Chen J
- 392 Three cancers in the renal pelvis, bladder, and colon: A case report
Chen J, Huang HY, Zhou HC, Liu LX, Kong CF, Zhou Q, Fei JM, Zhu YM, Liu H, Tang YC, Zhou CZ
- 399 Severe aconite poisoning successfully treated with veno-arterial extracorporeal membrane oxygenation: A case report
Kohara S, Kamijo Y, Kyan R, Okada I, Hasegawa E, Yamada S, Imai K, Kaizaki-Mitsumoto A, Numazawa S
- 405 Chemotherapy combined with bevacizumab for small cell lung cancer with brain metastases: A case report
Yang HY, Xia YQ, Hou YJ, Xue P, Zhu SJ, Lu DR
- 412 Diagnostic challenges and individualized treatment of cervical adenocarcinoma metastases to the breast: A case report
Akers A, Read S, Feldman J, Gooden C, English DP
- 418 Subsequent bilateral acute carpal tunnel syndrome due to tophaceous infiltration: A case report
Yeoh SC, Wu WT, Shih JT, Su WC, Yeh KT

- 425 Uniportal video-assisted thoracoscopic fissureless right upper lobe anterior segmentectomy for inflammatory myofibroblastic tumor: A case report
Ahn S, Moon Y
- 431 Hybrid treatment of varied orthodontic appliances for a patient with skeletal class II and temporomandibular joint disorders: A case report and review of literature
Lu T, Mei L, Li BC, Huang ZW, Li H
- 443 Significant improvement after sensory tricks and trunk strength training for Parkinson's disease with antecollis and camptocormia: A case report
Wang JR, Hu Y
- 451 Granulomatous mastitis in a 50-year-old male: A case report and review of literature
Cui LY, Sun CP, Li YY, Liu S
- 460 Double-chambered left ventricle with a thrombus in an asymptomatic patient: A case report
Kim N, Yang IH, Hwang HJ, Sohn IS

ABOUT COVER

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WJCC mainly publishes articles reporting research results and findings obtained in the field of clinical medicine and covering a wide range of topics, including case control studies, retrospective cohort studies, retrospective studies, clinical trials studies, observational studies, prospective studies, randomized controlled trials, randomized clinical trials, systematic reviews, meta-analysis, and case reports.

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Asymptomatic low-grade appendiceal mucinous neoplasm: A case report

Min-Quan Yao, Yu-Peng Jiang, Yuan-Yu Wang, Yi-Ping Mou, Jin-Xing Fan

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Abstract

BACKGROUND

Low-grade appendiceal neoplasms (LAMN) are characterized by low incidence and atypical clinical presentations, often leading to misdiagnosis as acute or chronic appendicitis before surgery. The primary diagnostic tool for LAMN is abdominal computed tomography (CT) imaging. Surgical resection remains the cornerstone of LAMN management, necessitating en bloc tumor excision to minimize the risk of iatrogenic rupture. Laparoscopy, known for its minimal invasiveness, reduced postoperative discomfort, and expedited recovery, is a safe and reliable approach for LAMN treatment. Despite the possibility of pseudomyxoma peritonei development, appendectomy and partial appendectomy generally result in negative tumor margins and favorable outcomes, which can be attributed to the disease's slow growth and lower malignancy.

CASE SUMMARY

A 71-year-old male patient was admitted to our hospital with a pelvic space-occupying lesion detected 1 mo prior. Physical examination showed a soft abdomen without tenderness or rebound and no palpable masses. No shifting dullness was noted, and digital rectal examination revealed no palpable mass. Enteroscopy revealed a raised, smooth-surfaced mass measuring 3.0 cm in the cecum. Abdominal contrast-enhanced CT showed a markedly thickened and dilated appendix with visible cystic shadows. Laparoscopic surgery was performed and revealed a significantly dilated appendix, leading to laparoscopic resection of the appendix and part of the cecum. Post-surgical pathologic analysis confirmed LAMN. The patient received symptomatic and supportive post-operative care and was discharged on postoperative day 4 without complications such as abdominal bleed-

ing, intestinal obstruction, or incision infection. No tumor recurrence was observed during a 7-mo follow-up period.

CONCLUSION

LAMN is a rare disease that lacks specific clinical manifestations. Abdominal CT plays a crucial role in diagnosing LAMN, and laparoscopic surgery is a safe and effective diagnostic and therapeutic approach.

Key Words: Low-grade appendiceal mucinous neoplasm; Pseudomyxoma peritonei; Computed tomography; Laparoscopy; appendectomy; Case report

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Core Tip: This study presents a case of asymptomatic low-grade appendiceal neoplasm (LAMN), highlighting the crucial role of abdominal computed tomography in its diagnosis. The early identification and treatment of LAMN are essential for mitigating the risk of pseudomyxoma peritonei and enhancing patient outcomes. Laparoscopic surgery is demonstrated to be a safe and effective diagnostic and therapeutic approach.

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INTRODUCTION

Appendiceal mucinous neoplasm (AMN) is a rare disease detected in less than 0.3% of appendectomy specimens and accounts for approximately 1% of gastrointestinal tumors[1]. In 2016, the Peritoneal Surface Oncology Group International classified AMNs into low-grade appendiceal neoplasms (LAMN), high-grade appendiceal neoplasms, mucinous adenocarcinoma, and goblet cell carcinoid[2]. LAMN is the most common pathological AMN subtype. LAMN is defined as mucinous neoplasms with low-grade cytology and any of the following: loss of muscularis mucosae, submucosal fibrosis, “pushing” infiltration (bulging or diverticular-like growth), undulating or flattened epithelial growth, rupture of the appendix, and/or mucinous cells and/or mucinous tissue outside the appendix[2]. LAMN has a low incidence and is more prevalent in adults and females. It is often incidentally detected during appendectomy[3].

On April 15, 2023, a patient was admitted to our hospital because of a pelvic space-occupying lesion. The lesion was surgically resected on April 18, 2023, and postoperatively confirmed as LAMN. Here, we describe this case, along with a literature review.

CASE PRESENTATION

Chief complaints

A 71-year-old male patient was admitted to the hospital because of a pelvic space-occupying lesion detected 1 mo previously.

History of present illness

Examination revealed a cystic space-occupying lesion in the lower right abdomen. The patient had no fever, nausea/vomiting, abdominal pain, abdominal distension, hematemesis, or black stools.

History of past illness

The patient had a 10-year history of hypertension that was controlled with 80 mg/12.5 mg valsartan and hydrochlorothiazide tablets once daily.

Personal and family history

He had a smoking history of approximately 20 cigarettes daily for 30 years and reported no alcohol consumption. There was no significant family medical history.

Physical examination

The patient was alert and oriented, and his mental status was unremarkable. There were no signs of enlarged or swollen superficial lymph nodes. Cardiac and pulmonary physical examination showed no apparent signs of abnormality. The



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Figure 1 Enteroscopy. A raised mass with a smooth surface measuring 3.0 cm was observed in the cecum.

patient's abdomen was soft, without any tenderness or rebound, and there were no palpable masses. Physical examination revealed no shifting dullness, and the patient's bowel sounds were approximately four times per minute. There was no palpable mass on digital rectal examination.

Laboratory examinations

Laboratory tests following admission showed a white blood cell count of $6.0 \times 10^9/L$, red blood cell count of $4.78 \times 10^{12}/L$, hemoglobin of 14.8 g/dL, platelet count of $234.0 \times 10^9/L$, and C-reactive protein concentration of 11.5 mg/L. The result of the fecal occult blood test was negative. Detection of tumor markers showed the following results: carcinoembryonic antigen, 10.83 ng/mL; CA72-4, 1.1 U/mL; and CA199, 5.1 kU/L.

Imaging examinations

A raised mass with a smooth surface measuring 3.0 cm was observed in the cecum during enteroscopy (Figure 1). Abdominal contrast-enhanced computed tomography (CT) showed a markedly thickened and dilated appendix with visible cystic shadows. The appendix wall showed a slight enhancement following contrast enhancement (Figure 2).

FINAL DIAGNOSIS

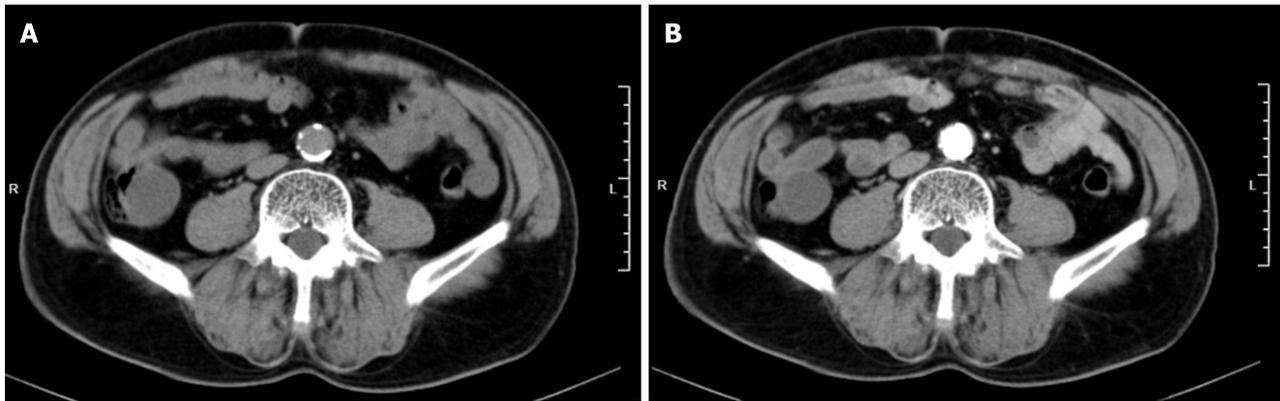
The final diagnosis of LAMN was pathologically confirmed.

TREATMENT

On the 3rd d of admission, laparoscopic surgery was performed and revealed a significantly dilated appendix (Figure 3A). A linear cutter/stapler was used to laparoscopically resect the appendix and portions of the cecum (Figure 3B). The appendix was 12 cm in length and 4 cm in width (Figure 4). Postoperative pathology confirmed that the tumor had invaded the entire appendix with mucus accumulation in the apical part of the appendix and the sub-plasma layer of the appendiceal body. The resection margins were negative (Figure 5). After surgery, the patient received symptomatic and supportive care, including treatment with cefuroxime 1.5 g twice daily for 2 d for preventing bacterial infections and nutrition therapy.

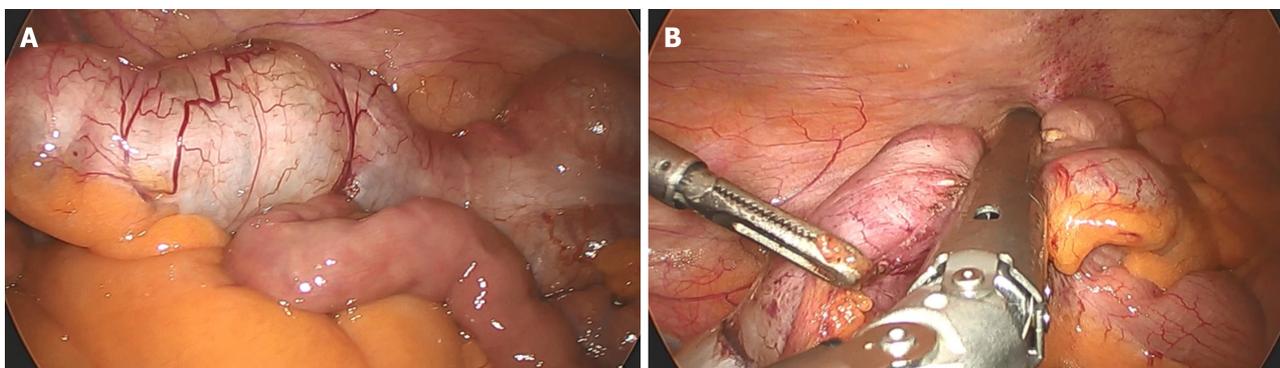
OUTCOME AND FOLLOW-UP

The patient was discharged from our hospital on postoperative day 4 without experiencing any complications such as abdominal bleeding, intestinal obstruction, or incision infection. Routine blood tests, biochemical analyses, tumor marker assessments, and chest and abdominal CT scans were performed in an outpatient setting on November 20, 2023, revealing no evidence of recurrence.



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Figure 2 Abdominal computed tomography. A: Markedly thickened and dilated appendix with visible cystic shadows; B: Appendix wall showed a slight enhancement following the contrast enhancement procedure.



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Figure 3 Intraoperative view. A: Significantly dilated appendix; B: Linear cutter/stapler was used to laparoscopically resect the appendix and portions of the cecum.

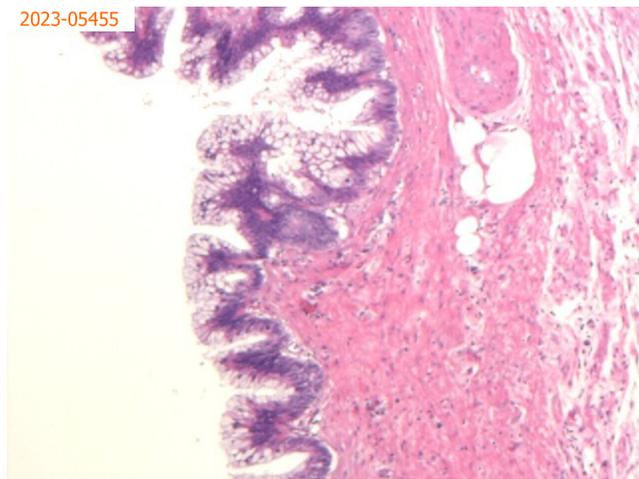


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Figure 4 Appendix. A: 12 cm in length; B: 4 cm in width.

DISCUSSION

Diagnosing LAMN is challenging because of its low incidence rate, atypical clinical features, lack of specific clinical symptoms, and absence of a specific tumor marker. The primary symptom of LAMN is pain in the lower right abdomen due to dilatation of the appendiceal lumen resulting from early-stage tumor growth. This may be mistaken for acute or chronic appendicitis before surgery[4]. Additional symptoms of LAMN include weight loss, nausea/vomiting, decreased appetite, and changes in bowel habits, which can lead to intestinal obstruction, intussusception, gastrointestinal he-



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Figure 5 Postoperative pathology. The tumor had invaded the entire appendix with mucus accumulation in the apical part of the appendix and the sub-plasma layer of the appendiceal body. The resection margins were negative (hematoxylin-eosin, × 200).

morrhage, ureteral compression, and secondary infections and necrosis/hemorrhage from the tumor in severe cases[5]. Approximately 25% of LAMN patients are asymptomatic, and the disease is often detected during health check-ups or surgical procedures[6].

Tumor rupture is the most serious and frequent complication of LAMN. It can cause the spread of tumor cells and mucinous ascites into the intraperitoneal cavity, which can result in pseudomyxoma peritonei (PMP)[7], a severe clinical syndrome with high morbidity and mortality. Therefore, early identification and treatment of LAMN is crucial in mitigating the risk of PMP and enhancing patient outcomes.

Imaging is crucial for both LAMN diagnosis and treatment. AMNs are characterized by mucus accumulation and the "onion skin" sign on ultrasound[8]. LAMN is most commonly diagnosed through CT imaging, as it reveals two common features of LAMN: cystic dilation of the appendiceal lumen and irregular thickening of the appendiceal wall. Wall calcification may accompany the latter. Mucinous tissue typically does not reach the cecal lumen because of blockage in the appendiceal lumen. Colonoscopy may reveal a smooth elevated mass originating from the opening of the appendix, without any visible mucinous tumor tissue.

LAMN has the potential to become malignant, and rupture of the appendiceal wall can lead to PMP. Surgical resection is the primary treatment for LAMN, during which the tumor must be excised en bloc to minimize the risk of iatrogenic rupture. Ideally, LAMN lesions at different sites should be completely resected, ensuring negative surgical margins. Regional lymphadenectomy is not typically required[9]. For LAMNs found in the distal appendix, removal of the appendix may be sufficient. If the tumor involves the appendiceal root or adheres to the cecum, removal of part of the cecum may be required. Finally, for large and malignant tumors, ileocolic resection or right hemicolectomy will be required[10]. Intraoperative frozen section pathology can be useful in identifying the nature of the tumor and resection margins. During open surgery, the tumor can be placed outside the body and surrounded by gauze to prevent tumor cell dissemination, thus reducing surgical risks. Laparoscopy offers advantages over open surgery as it can reduce stress responses, relieve postoperative pain, and reduce length of postoperative stay[11,12]. According to Singh *et al*[13], laparoscopic appendectomy using a specimen retrieval bag was a safe treatment option for LAMN when surgery was performed with low pneumoperitoneum pressure. Despite the possibility of PMP development in patients with LAMN, appendectomy and partial appendectomy often produce negative tumor margins and yield good outcomes because of the slow and less malignant nature of the disease process.

CONCLUSION

LAMN is a rare disease that lacks specific clinical manifestations and is usually identified incidentally following appendectomy. Before undergoing an appendectomy, a thorough medical history and physical examination are necessary. Medical imaging plays a crucial role in diagnosing LAMN. Prompt abdominal CT and enteroscopy are crucial for LAMN diagnosis, and laparoscopic examination is a safe and effective diagnostic and therapeutic approach. Appendectomy and partial appendectomy usually result in favorable outcomes, including a possible cure.

FOOTNOTES

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content, read and approved the final manuscript, and take public responsibility for appropriate portions of the content and have agreed to be accountable for all aspects of the work.

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