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Case report

A rare case of a Low Grade Appendiceal Mucinous Neoplasm in a 46-year-old Middle Eastern female - A case report

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ABSTRACT

Introduction and importance: Mucinous cell neoplasia are rare and conceivably fatal causes of surgical abdomen. Adult manifestations can be obscure and ill-defined, yielding misdiagnoses and wasting precious time and effort to reach the correct one. Clinical awareness ought to be present once presented with a surgical abdomen in such a patient because this neoplasm may present with misleading presentations which mask the actual diagnosis and masquerade as a different one, and in turn, may result in performing a dissimilar treatment intervention. Rapid learnt surgical judgments must be taken and put into action to diminish the morbidity and mortality consequential to this pathology.

Case presentation: Our case is of a 46-year-old female, who was admitted into our hospital with a 2-month-history of general fatigue. Colicky abdominal pain and discomfort developed shortly prior to admission. Multi-Slice Computed Tomography (MSCT) scan exposed cystic formation in the terminal ileum. Exploratory laparotomy was warranted, in addition to performing a right hemicolectomy.

Clinical discussion: We treated our patient via open surgery and performing an appendectomy with right hemicolectomy in addition to the excision of several surrounding lymph nodes. Diagnosis is traditionally reached intraoperatively relying on gross morphology and postoperatively through histopathological analysis of the excised specimens.

Conclusion: Low Grade Appendiceal Mucinous Neoplasm is a scarce entity and varies critically in its manifestations, hence, it is existential to thoroughly study this kind of neoplasia, document it, and consider it, so that we can construct precise decisions to reach ideal results for patients who suffer from this neoplasm.

1. Introduction

Appendiceal primary tumors have an incidence rate of less than 2% of all excised samples of the appendix [1,5]. Primary tumors of the appendix are classified into the following types: lymphomas, mesenchymal cell neoplasms, and epithelial cell neoplasms. Appendiceal mucinous cell tumors are denoted under appendiceal epithelial cell tumors. This type of neoplasms repeatedly culminates in cystic dilation of the appendix. This transpires as a ramification of the buildup of materials which are gelatinous in composition. They have the nomenclature of mucoceles due to their shape and texture. Rokitansky in 1842 was the pioneer who depicted this type of tumors [2]. Mucinous cell tumors of the appendix are ubiquitous in merely 0.2%–0.3% of all the excised Appendiceal specimens [3].

The work has been reported in line with the SCARE criteria and the revised 2020 SCARE guidelines [4].

Abbreviations: MSCT, Multi-Slice Computed Tomography; IV, Intravenous; LAMN, Low Grade Appendiceal Mucinous Neoplasm; IBS, Irritable Bowel Syndrome.

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2. Presentation of case

2.1. Patient information

We present the case of a 46-year-old Middle Eastern female who is a known case of Hashimoto's Thyroiditis, who presented to the general surgery clinic with symptoms commencing 2 months prior to admission. Our patient informed us of suffering from general fatigue, tiredness, and overall "lack of energy". She didn't suffer from dizziness, syncope, weight loss, loss of appetite, night sweats, or fever. Moreover, she did not report any nausea, vomitus, or alternation of bowel habits. 10 days prior to admission, she experienced vague periumbilical abdominal pain unresponsive to over-the-counter analgesia. She was referred to our university hospital from an internal medicine clinic for the suspicion of a surgical cause behind her symptoms. No genitourinary symptoms were reported. She has negative family, drugs, and allergic histories. She is not a smoker nor an alcohol consumer. Her BMI is 23 kg/m^2.

2.2. Clinical findings

Physical examination demonstrated sinus tachycardia coincided by mild tachypnea. Apart from that, her other vital signs were within normal ranges. When inspecting her abdomen, it wasn't tumid. Nonetheless, it moved agonizingly with respiration. Upon superficial palpation, there was periumbilical and right iliac fossa guarding and mild discomfort. Upon deep palpation, no masses were felt. Upon auscultation, peristaltic sounds were exaggerated.

Laboratory investigations revealed anemia (Hb: 10.7 g/dl) and mild

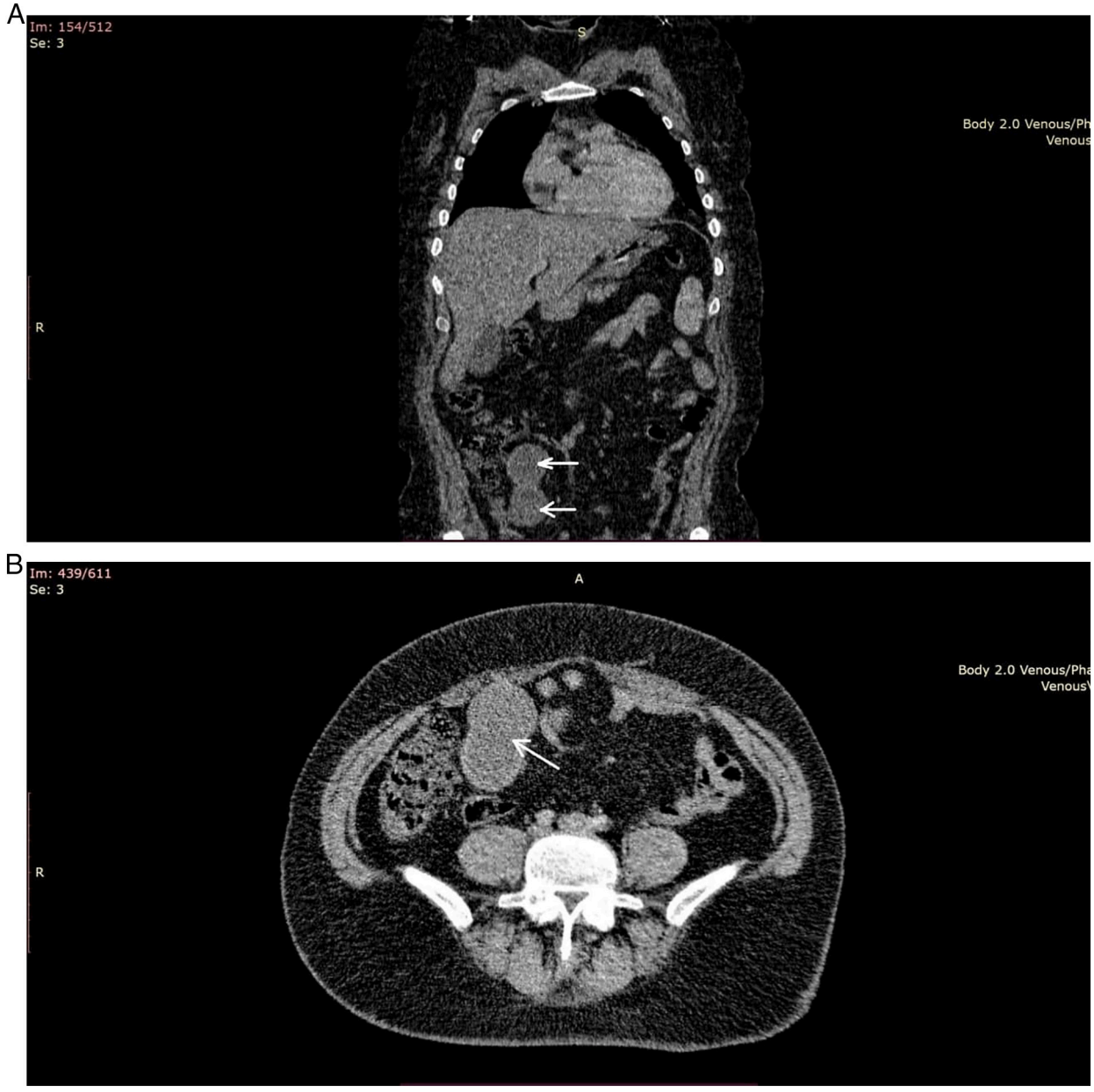

Fig. 1. A–B: MSCT scan of the abdomen and pelvis with coronal and sagittal views, white arrows in the respective views demonstrate a well-circumscribed, low-attenuation, spherical cystic mass, with internal homogeneous non-enhancing contents, slightly thickened enhancing wall with no mural calcification, and located anterior to the right psoas muscle contiguous with the base of the cecum. It measures approximately (8.5 × 5.5 cm).
No radiographic features for infectious etiologies, no surrounding fat stranding, no lymphadenopathy was noted, no free fluid is seen.
Findings are conforming with an appendiceal mass. Furthermore, we cannot rule-out other lesions or cystic neoplasms.

hypokalemia (Potassium was 3.31 mmol/l), her TSH was normal as the patient was being prepared for surgery (TSH: 2.3 miu/l). Tumor marker (CA 19–9: 0.60 u/ml) which was normal. The rest of the investigations yielded normal results.

2.3. Diagnostic assessment

Abdominal ultrasound demonstrated a lesion with hypoechoic and hyperechoic composition situated in the right iliac fossa and estimated to be measuring (9.5 × 6.5 cm). It is unrelated to the right ovary. Result most likely conforms with endometriosis or tuberculosis with necrotic nodes -according to the radiologist-.

Trans-vaginal ultrasound was performed to exclude any potential reproductive system pathology and it found the uterus and the ovaries to be free of abnormal findings. On the other hand, it confirmed the presence of a mass in the right iliac fossa to be correlated with CT imaging. Multi-Slice Computed Tomography (MSCT) scanning with coronal and sagittal views, white arrow in the respective views demonstrates a well-circumscribed, low-attenuation, spherical cystic mass, with internal homogeneous non-enhancing contents, slightly thickened enhancing wall with no mural calcification, located anterior to the right psoas muscle contiguous with the base of the cecum. Measuring approximately (8.5 × 5.5 cm).

No radiographic features for infectious etiologies, no surrounding fat stranding, no lymphadenopathy was noted, and no free fluid is seen.

Findings are conforming with an appendiceal mass. Furthermore, we cannot rule-out other lesions or cystic neoplasms (Fig. 1A-B).

Furthermore, the lungs, liver, and pancreas were scanned and were deemed free of any abnormal lesions.

To complete the radiological preoperative study, a thyroid ultrasound was performed and revealed a moderate enlargement with echogenic heterogeneity. Additionally, we marked the presence of numerous hyperechoic lesions conforming with the features of Hashimoto's Thyroiditis. No nodal lesions were noted.

Initial therapeutic approach included setting-up a large-bore intravenous (IV) cannula, IV fluid resuscitation, potassium levels replenishment via the administration of IV potassium chloride, preoperative IV antibiotics for surgical preparation, and a complete laboratory panel commencing with blood sampling and crossmatch for blood transfusion to replenish the Hemoglobin levels of the patient and to safeguard any surgical needs.

One notable obstacle, which altered the intervention modality, was the unavailability of a laparoscopic device in the hospital at the time of patient referral due to a technical malfunction. Furthermore, the patient is of a low socioeconomic status, thus, this disallowed her from seeking a medical facility with such a device.

2.4. Therapeutic intervention

An exploratory laparotomy was indicated on the basis of the provided clinical picture. The surgery was performed at our tertiary university hospital. It was achieved by a fifth-year senior general surgery resident with five years of experience, a PhD general surgery student with 7 years of experience, and by a general surgery professor with 35 years of general surgery experience. The procedure was seen through under general anesthesia with no anesthetic complications. Laparotomy had emphasized the radiological findings of the MSCT scan. Via exploration, a well-circumscribed cystic mass at the region of the ileo-cecal junction, overwhelming the appendix and extending over the cecum, was discovered, roughly measuring (10 × 12 cm). No gross pathological appearance of surrounding lymph nodes was seen, no ascites, or gross necrotic material was seen. The rest of the abdomen including the omentums, peritoneum, internal organs including the liver, ovaries, pancreas, and stomach were explored and no lesions or seeding from the tumor were grossly noted. A right hemicolectomy was therefore warranted in addition to excising 10 cm from the terminal ileum. Afterwards, a side-to-side ileo-colonic anastomosis was accomplished via stapler.

Intraoperative photographs point-out the terminal ileum, cecum, and the neoplastic masses in the appendiceal region (Fig. 2).

Intraoperative post-resection photographs depict the excised specimens which were the ascending colon, cecum, the appendix along with the masses arising from it, and approximately 10 cm from the ileum (Fig. 3) All of the excised materials were sent for histopathological analysis. Histopathological report revealed a cystic formation in the site of the cecum and appendix measures (11 × 5.5 × 3.2 cm), containing a gelatinous, mucinous yellow substance, with white areas. Diagnosis was revealed to be a mucinous cell neoplasm of the appendix. Findings further suggest Low Grade Appendiceal Mucinous Neoplasm (LAMN). Acellular mucinous pools seen within the submucosal tissue and dissect through muscle layers. No serosal involvement was seen. Mucinous pools within submitted sections were noted (Fig. 4) The lining mucous-secreting epithelium was noted (Fig. 5) The distal and proximal lines of resection are free of pathological involvement. All the isolated lymph nodes are free of invasion by neoplastic cells (Fig. 6) Our patient underwent complete postoperative recovery and therefore, was discharged home within 6 days of the operation.

She was informed of the nature of her illness and the details of histopathological results. Moreover, she was given a comparative list of several lifestyle modifications which may pave the path to a full recovery, such as regular sterile wound dressings by a professional, analgesics for pain management, a medical prescription of postoperative antibiotics, and a meticulous follow-up regimen. A follow-up protocol was set in motion in the outpatient settings for three months so far and is still being done. She has had regular visits at the general surgery clinic to receive surgical evaluations via clinical examination and postoperative ultrasound imaging. Follow-up regimens also included laboratory investigations and thorough physical examination. She is currently deemed free of any relapse or recurrence of any lesions.

3. Discussion

Appendiceal neoplasias are pervasive in approximately only 0.4% to 1% of all the tumors of the gastrointestinal tract [6,7], of all these excised appendiceal specimens, 0.7% to 1.7% is the incidence rate of appendiceal tumors discovered incidentally as a result of the respective histopathological analysis [6–10]; Amongst the previous percentage of specimens, 0.2% to 0.3% is the incidence rate of appendiceal mucinous cell tumors [5,11,12].

Several distinctive neoplastic etiologies reflecting these tumors are noted, such as mucinous adenoma, mucinous adenocarcinoma, carcinoid tumors, cystadenocarcinoma, Non-Hodgkin's Lymphoma, adenocarcinoid, and Low Grade Appendiceal Mucinous Cell Neoplasms (LAMN) [7].

The prognosis of such cases will heavily rely on the neoplastic grade and the extent of neoplastic invasion to the tumoral surroundings. LAMN is the nomenclature which is utilized to depict a primary tumor with epithelial dysplasia. This neoplasm generates copious amounts of mucin and has a distinguished characteristic attribute of growing in an expansive manner which shows a “pushing” border. The result of such expansion might be the obliteration of the muscular elements of the appendiceal wall in addition to mural fibrosis [9,29].

LAMNs generally do not possess the capability of vivid epithelial infiltration and invasion of the walls of the appendix. Instead, they are predominantly limited and surrounded by the appendiceal muscularis propria layer. In some instances, we might witness the penetration of mucin through the wall of the appendix. However, the conglomeration of mucin is acellular in nature. With regards to gender favorability, males and females are at an interchangeable comprehensive risk to manifest a type of appendiceal mucinous cell neoplasm [7,18]. Nevertheless, a scarce number of articles demonstrated that these tumors may favor occurring in females more than males [13–17]. Amongst the

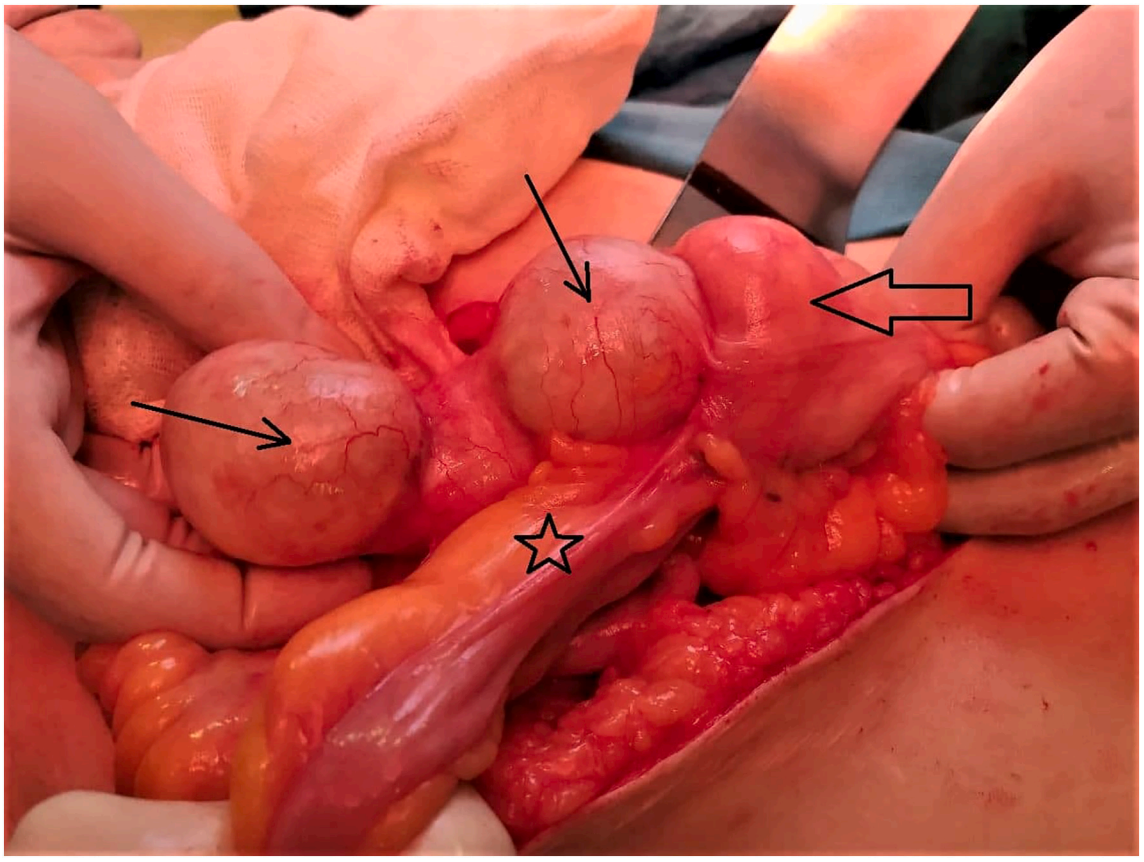

Fig. 2. Intraoperative image prior to tumor excision. Hollowed arrow points towards the cecum. The star identifies the terminal ileum. The two remaining arrows mark the identified neoplasm at the appendiceal location.

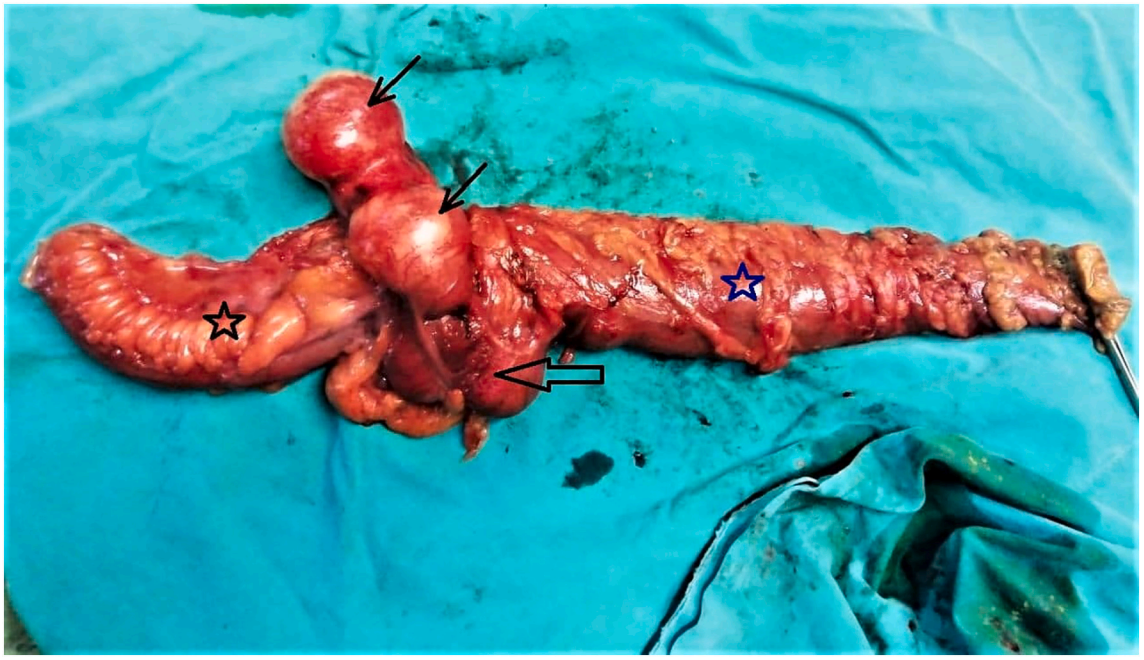

Fig. 3. Intraoperative image post-resection of the tumor. Black star depicts the excised portion of the terminal ileum. Hollow Arrow points to the cecum. The two adjacent, black-filled arrows point towards the mucinous cell neoplasm. Blue star depicts the excised ascending colon. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

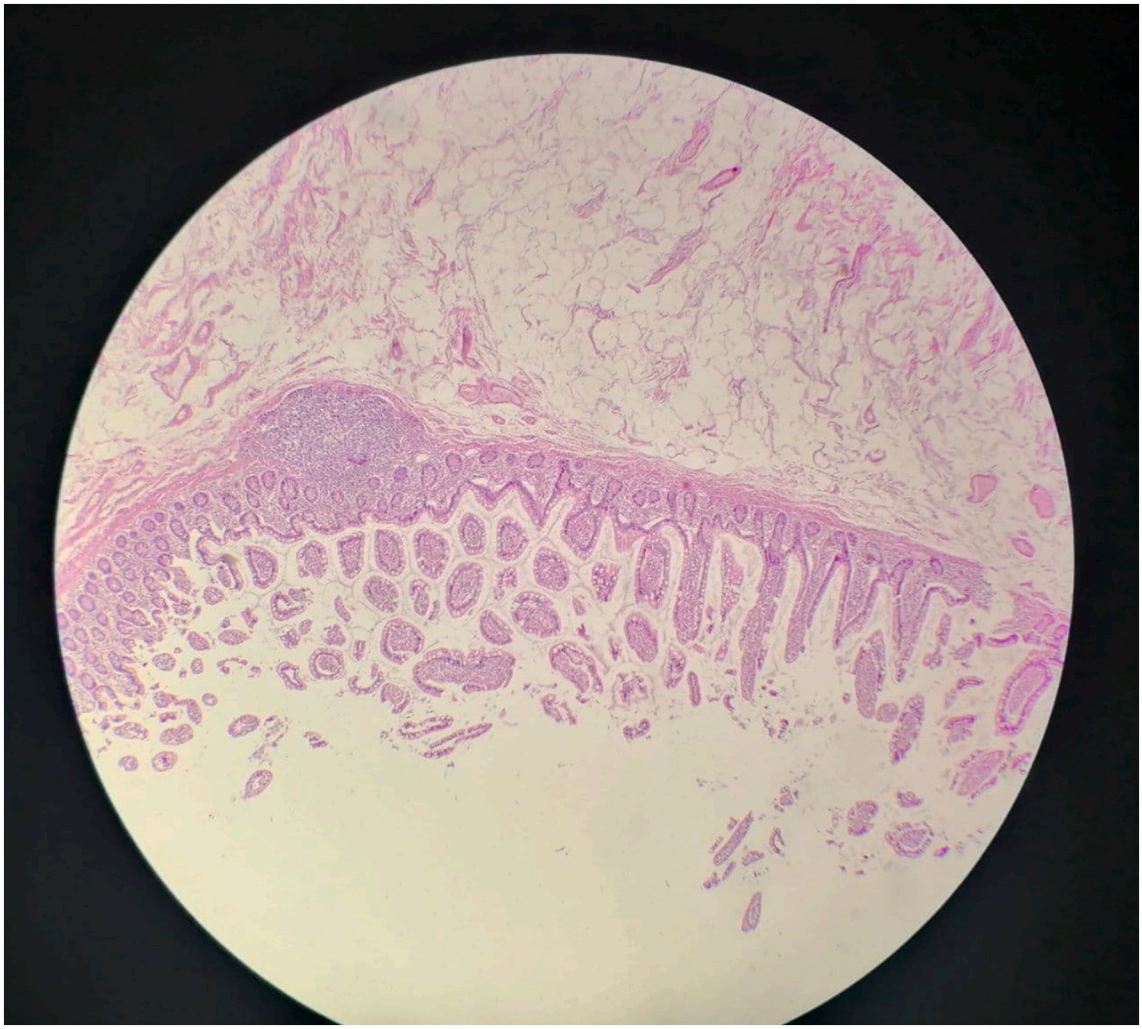

Fig. 4. Histopathology depicts a cystic formation in the site of the cecum and appendix containing a gelatinous, mucinous yellow substance, with white areas, suggestive of Low Grade Appendiceal Mucinous Cell Neoplasm (LAMN). Acellular mucinous pools within the submucosal tissue and dissect through muscle layers. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

studied samples, this type of tumors is found in individuals older than 50 years [5,15,17–19].

Acute appendicitis is the established clinical diagnosis in almost 30–50% of cases where appendiceal tumors are the actual diagnosis. This misdiagnosis occurs because of the non-specific signs and symptoms of said tumors of the appendix. In other occasions, these tumors can otherwise masquerade as an asymptomatic abdominal mass demarcated upon palpation, lower gastrointestinal system hemorrhage, intussusception, a vivid increase in abdominal wall contour when a mucocele ruptures inside the abdomen, or as an incidental radiological finding [11,20–22].

Seldom, these tumors can manifest as hernias such as inguinal or umbilical hernias [8,9] or as irritable bowel syndrome (IBS) [23] or as chronic non-specific abdominal pain [24].

With regards to appendiceal adenocarcinomas, the chief presentation is pain and tenderness in the right iliac fossa, elevation in white blood cell count, episodes of pyrexia, and loss of appetite. This can morph the illness as a case of acute appendicitis [6,7,9,25,26].

Sporadically, this pathology can evince itself as a palpable abdominal or pelvic mass [6,9,25], increased contour of the abdomen, mild dyspnea and tachycardia, general fatigue, weight gain [8], or simply as a case of acute surgical abdomen [26].

The gold standard of therapeutic approaches is surgical resection. Nevertheless, the depth and extent of surgical resection relies ponderously on the grade of tumor and the size of the mucocele. Additionally, it rests on whether the mucocele has perforated or not [27].

For further depiction of the guidelines available, any mucocele with a size larger than 2 cm, the operation of choice is a right hemicolectomy [5,22]. As for mucoceles with a size smaller than 2 cm, only an appendectomy is indicated [22].

In broader terms, surgery via the open route yields better results than laparoscopic surgery. This is due to the lesser incidence of mucocele perforation with open surgery than with laparoscopic intervention. This perforation can lead to the development of peritoneal pseudomyxoma [28]. Nonetheless, some studies suggested that there might be a parallel risk for the development of mucocele perforation when comparing open surgery with laparoscopic surgery [28].

4. Conclusion

To sum up, the nebulous and imprecise clinical manifestations of LAMN of the appendix mandate that we invest a significant degree of clinical focus on this noteworthy pathology.

It can easily profess as another form of neoplasms and mimic diverse pathologies. Misdiagnosis can be the ramification, and this may denote several different clinical approaches. Furthermore, we are bound to thoroughly consider this distinctive type of appendiceal neoplasm when we are presented with a female patient suffering from symptoms

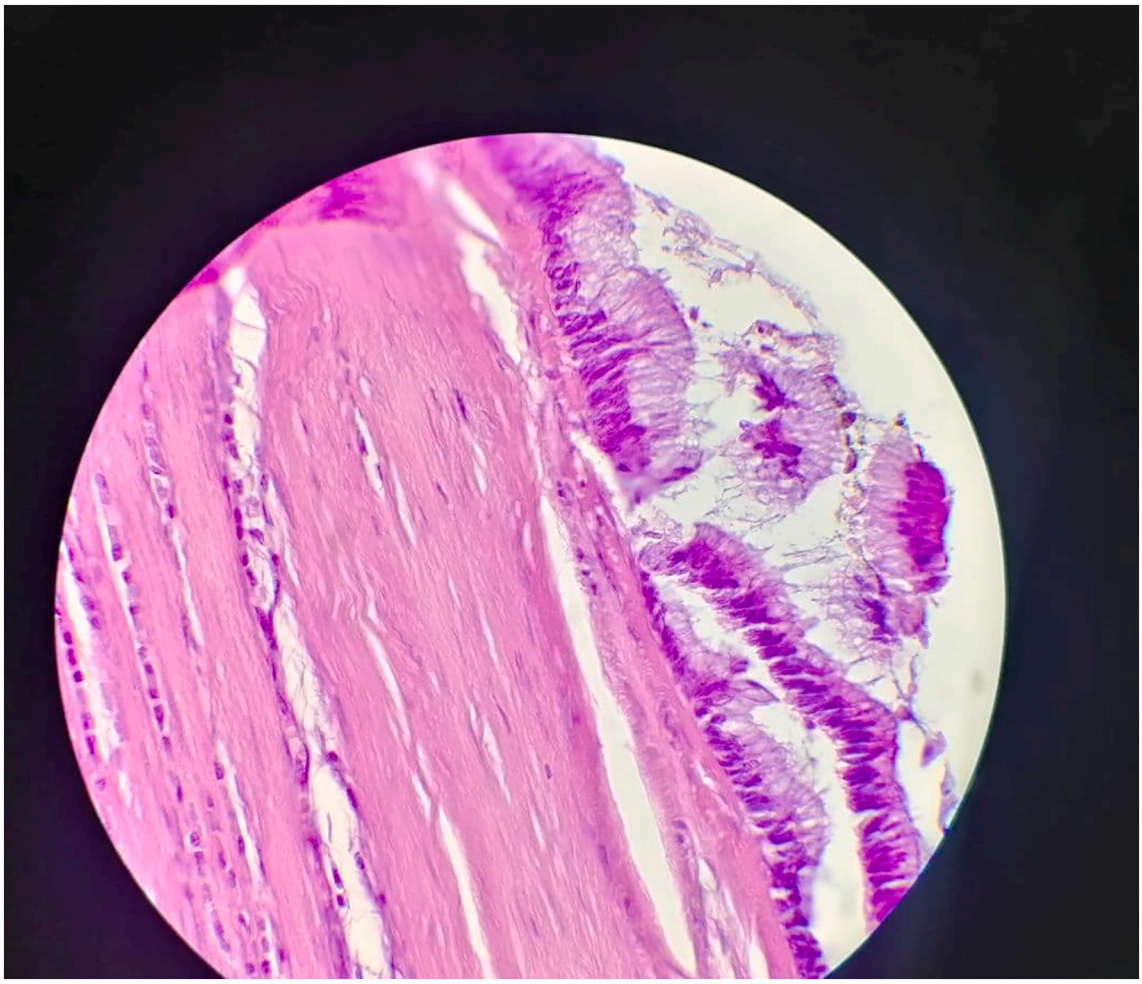

Fig. 5. Histopathology depicts the lining mucous-secreting epithelium.

resembling those of our patient. Her gender and age group are major indicators to several different pathologies.

Nevertheless, making time-efficient surgical judgments will assist in saving countless lives, spare the utilization of unnecessary resources, save time for the physician and the patient, and yield in satisfactory outcomes.

The morbidity and mortality of this neoplasm will be immensely limited when we diagnose it correctly and implement the correct management when presented with the opportunity.

Lastly, documentation is the corner stone on which we rely in surgery to perform studies and build treatment protocols for patients. This case can aid in documenting such a rare occurrence so that further studies can be established with regards to this neoplasm.

Abbreviations

MSCT	Multi-Slice Computed Tomography
IV	intravenous
LAMN	Low Grade Appendiceal Mucinous Neoplasm
IBS	irritable bowel syndrome

Ethics approval and consent to participate

Institutional review board approval is not required for deidentified single case reports or histories based on institutional policies.

Consent of patient

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Availability of data and materials

The datasets generated during and/or analyzed during the current study are not publicly available because the Data were obtained from the hospital computer-based in-house system. Data are available from the corresponding author upon reasonable request.

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Author contribution

OA, RA: Who wrote, original drafted, edited, visualized, validated, and literature reviewed the manuscript.

AA: Review of the manuscript and assistance in the radiological analysis of the imaging studies.

MA: PhD General Surgery student who assisted in the performance and supervision of the surgery. Supervision and review of the

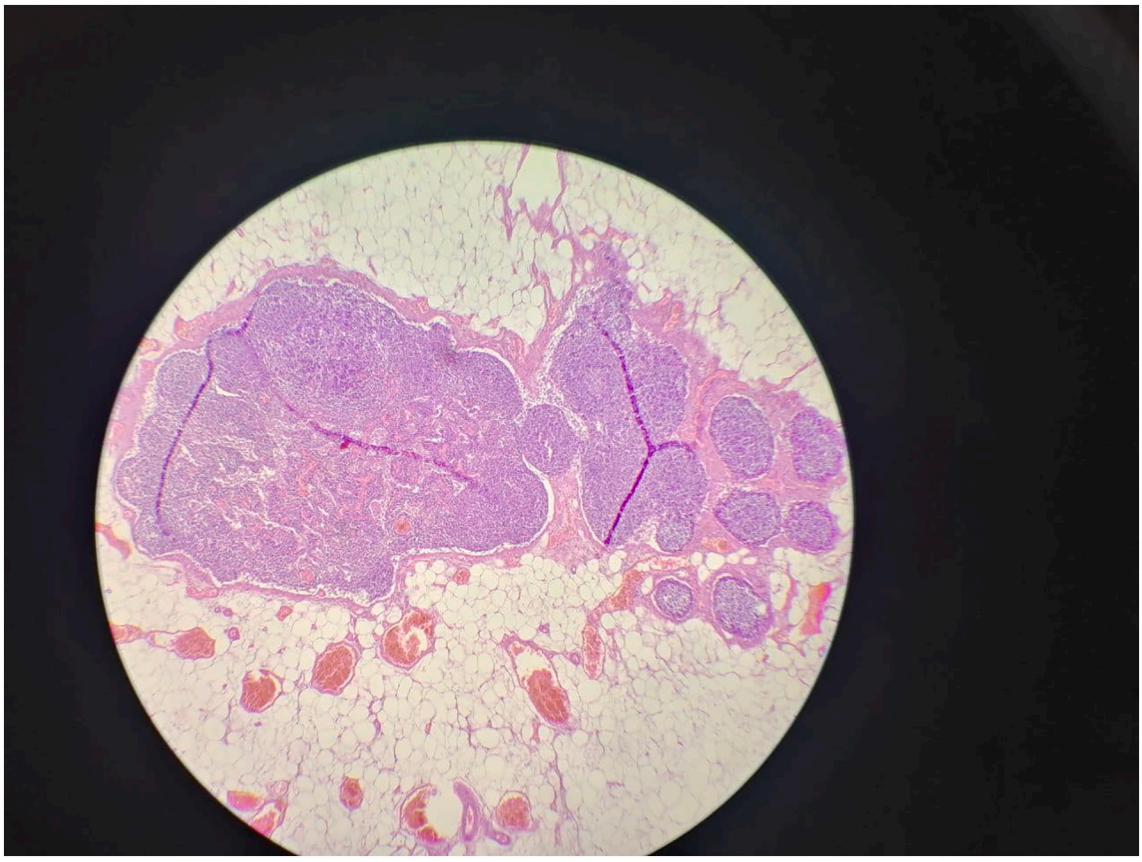

Fig. 6. Histopathology depicts a microscopic view of the excised surrounding lymph nodes with no evidence of neoplastic involvement.

manuscript.

FA: General Surgery senior resident who was the first assistant in the surgery. Supervision, project administration, and review of the manuscript.

HH: MD, PhD, General Surgery Professor who performed and supervised the operation. Supervision and review of the manuscript.

OA: Conceptualization, resources, and the corresponding author who submitted the paper for publication.

All authors read and approved the final manuscript.

Research registration

N/A.

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Declaration of competing interest

The authors declare that they have no competing interests.

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Pathology Department at Al Assad University Hospital, Damascus, Syria.

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