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Case Report

Massive appendiceal mucinous neoplasm diagnosed after abdominal trauma: Pitfalls in imaging and diagnosis ☆☆☆

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ABSTRACT

Low grade appendiceal mucinous neoplasm (LAMN) is a rare tumor presenting typically with symptoms of acute appendicitis. A 33-year-old gentleman presented with abdominal fullness and discomfort secondary to abdominal trauma obtained during boxing training. Investigations with magnetic resonance imaging (MRI), ultrasound, and colonoscopy revealed a large cystic lesion in the ascending colon mesentery with unclear continuation with the appendix. There were no obvious features of malignancy. Laparoscopy revealed a large, firm mass connected to the appendix with enlarged lymph nodes, and the patient underwent radical resection with a right hemicolectomy. Histopathology revealed complete excision of LAMN with no lymphatic involvement. Post traumatic intra-abdominal cystic lesions pose a diagnostic challenge, and neoplastic lesions cannot be ruled out with imaging alone, even with benign appearances. Careful surgical excision and histopathological diagnosis is the only definitive method of ruling-out malignancy.

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Introduction

Appendiceal mucinous neoplasms account for less than 1% of all appendiceal specimens on histology [1]. They are a rare entity – with only 1 to 2,000 cases diagnosed in the United States each year [2]. These lesions are usually diagnosed in patients

in their fifth decade and are more prevalent in females [3]. Additionally they are more likely to be found in patients presenting with complicated appendicitis [4]. Tumor markers may be elevated in these cases, however they are nonspecific [5].

The initial presentation of appendiceal mucinous neoplasm masquerades as that of acute appendicitis [6]. Mucoceles can also be associated with gastrointestinal bleeding

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if they intussuscept or obstruct due to mass effect [7,8]. Although benign mucinous lesions rarely rupture, low-grade appendiceal mucinous neoplasms (LAMNs), high-grade appendiceal mucinous neoplasms (HAMNs) or mucinous adenocarcinomas may do so, leading to pseudomyxoma peritonei [9].

There is a paucity of reports regarding post-traumatic cystic lesions, although mesenteric cysts post trauma has been reported [10]. Most intra-abdominal cysts including mesenteric cysts can have variable features on imaging [11]. Differentiating benign and neoplastic lesions on imaging is clinical challenge, including diagnosing abdominal cystic lesions. We hereby describe a case of a low-grade mucinous neoplasm mimicking a mesenteric cyst on imaging after abdominal trauma.

Case report

A 33-year-old man was referred to our surgical center with a history of lower abdominal pain and fullness. These symptoms were precipitated by an episode of abdominal trauma during a boxing training session. Upon investigation a palpable mass was felt in the right lower quadrant. He had no significant past medical history or surgical history.

An abdominal MRI was conducted, revealing a $58 \times 52 \times 109$ mm intraperitoneal cystic lesion in the right mid to lower abdomen. The inferior margin of the lesion was in close proximity to the caecal pole, thin walled and entirely simple. The interior of the lesion demonstrated T2 hyperintensity but minimal T1 hyperintensity on weighted imaging. The appendix was not identified separate to this lesion as displayed in Figs. 1 and 2. Differential diagnosis included a mesenteric cyst, duplication cyst, and given the appendix was not separately visualized, an appendiceal mucinous lesion.

To further characterize the mass, an abdominal ultrasound was performed, describing a well-circumscribed lesion in the right mid-lower abdomen, distinct from the small bowel and appendix, with no apparent communication. Additionally, a moderate amount of debris and increased echogenicity was seen within the lesion with no associated intrinsic vascularity as seen in Fig. 3.

Colonoscopy was performed with visualization to the terminal ileum. No abnormalities were visualized and the appendiceal opening appeared normal. Tumor markers including CEA, Ca-19-9, and Ca-125 were normal.

The patient underwent a laparoscopy after consideration of their symptoms, investigation results and taking their preference into account. Considering investigations to date, ongoing symptomatology and patient preference, a laparoscopy was performed. A large inflammatory abdominal lesion with associated enlarged lymph nodes was visualized within the mesentery of the ascending colon, shown in Figs. 4 and 5. The appendix was not visualized. A right hemicolectomy with primary anastomosis was performed. Histopathology revealed a completely excised low-grade appendiceal mucinous neoplasm with fifteen negative lymph nodes.

The patient had an unremarkable long term postoperative course, with full return to baseline functioning.



Fig. 1 – Coronal T-2 weighted MRI abdomen-pelvis demonstrating a large tubular lesion with T-2 hyperintensity situated in the right lower abdomen within the mesentery of the colon.

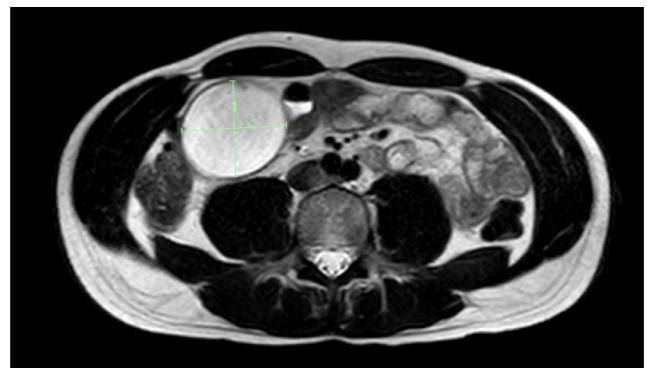


Fig. 2 – Axial T-2 weighted MRI abdomen-pelvis demonstrating a tubular lesion with T-2 hyperintensity situated adjacent to the caecum / ascending colon.

Discussion

Presented is an interesting case of an incidental finding of LAMN masquerading as a mesenteric cyst on imaging after a history of abdominal trauma. LAMN are rare, consisting of 8%–10% of all appendiceal tumors [12]. Typically, they present with acute appendicitis symptoms of right lower quadrant pain, which is secondary to distension of the appendix by mucin. These tumors rarely metastasise outside of the peritoneal cavity [6,13].

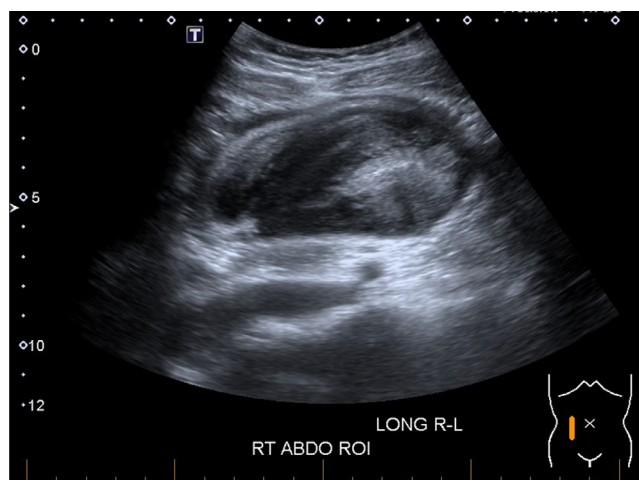


Fig. 3 – Longitudinal abdominal ultrasound of right lower abdominal lesion with moderate amount of debris seen within. No surrounding free fluid is observed.

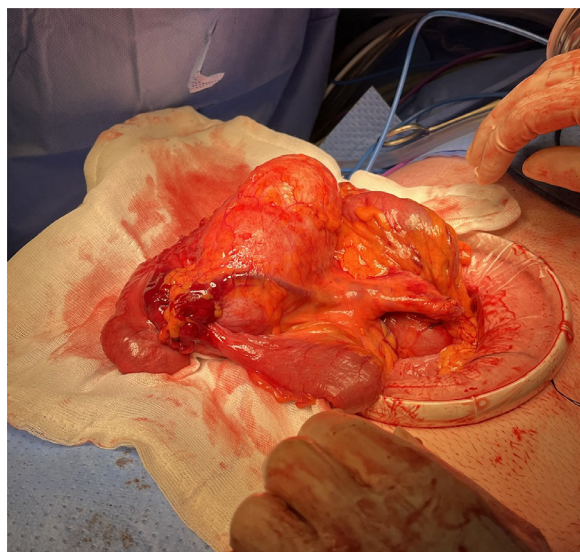


Fig. 4 – Appendiceal mass situated within bowel mesentery exteriorised intraoperatively.

Post traumatic mesenteric cysts have previously been reported in literature [10]. Falidas *et al.* reports a case of mesenteric cyst after abdominal trauma and hypothesized the cyst to be a consequence of mesenteric lymphangitic rupture or a haematoma post absorption with cystic degeneration [10,14]. However, in our case on MRI the cystic mass could not be distinguished from the appendix and therefore a mucinous neoplasm could not be ruled out, necessitating further pathology and radiology workup.

An alternative differential is that of a duplication cyst, a rare congenital lesion with an incidence of 1 in 4500 births and male predisposition [15]. Colonic or hindgut duplication cysts are rare in comparison to foregut and midgut distribution [16]. Most present symptomatically before 2 years of age, and characteristically appear with a 5-layered cyst wall with

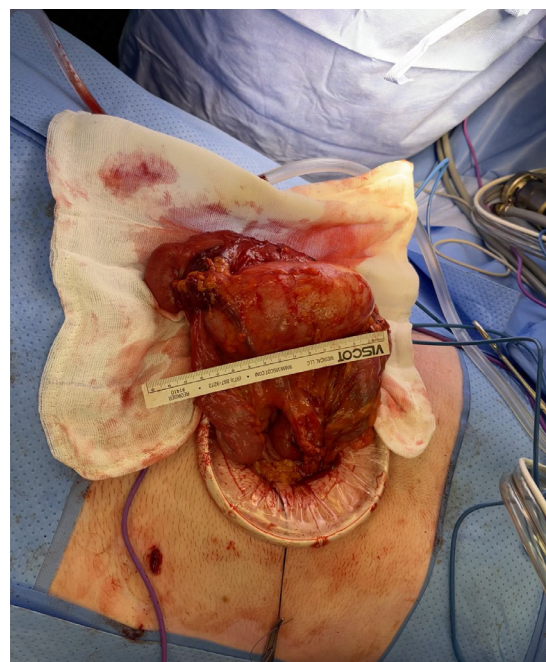


Fig. 5 – Scale clinical image of appendiceal mass situated within mesentery.

alternating hyperechoic and hypoechoic layers on ultrasound evaluation [17]. Although unlikely, we hypothesized our patient may have possessed an asymptomatic duplication cyst which was aggravated and enlarged secondary to abdominal trauma. The definitive treatment and diagnosis of such duplication cysts is surgical excision [17].

Appendiceal mucinous tumors typically have nonspecific laboratory findings but can present with elevated tumor markers such as carcinoembryonic antigen (CEA), Ca-125, and CA19-9. It is reported that the majority of patients with high grade tumors will present with elevated tumor markers, and the level of elevation is positively correlated with treatment outcomes [5]. Endoscopic findings of appendiceal mucinous neoplasms include a smooth indentation of caecal lumen, or the pathognomonic “volcano sign”, a mount like elevation surrounding the appendiceal orifice with inflammatory exudate extruding [18,19].

Radiological investigation plays an important role in the diagnosis of appendiceal mucinous lesions but lacks the ability to distinguish between neoplastic and non-neoplastic lesions. In saying that, there are characteristics which raise clinician's index of suspicion regarding neoplastic lesions, such as larger size and wall thickening or irregularities [20]. Computerised tomography (CT) is often the imaging modality of choice, however MRI can be superior in the detection of peritoneal disease [21]. MRI findings of AMN is often a T2 hyperintense lesion, with T1 hyperintensity dependent on the amount of contained mucin lesion [22]. Ultrasound is also utilized and has pathognomonic signs such as the “onion skin sign” [23].

The treatment of mucinous neoplasms is surgical resection, regardless of grade, due to the lack of reliable criteria to exclude malignant lesions on imaging [24]. The extent of surgery is dependent on the spread of disease; in localized

disease an appendectomy is often sufficient, however if the base of appendix is involved a partial caecectomy or even right hemicolectomy, as in this case, can be performed to ensure a sufficient oncological resection [13]. Ruptured lesions warrant extensive lavage or even formal cytoreductive surgery [25]. In our case, given the macroscopically enlarged lymph nodes we elected to perform a right hemicolectomy for oncological resection with a sufficient lymph node yield.

In summary, the cystic lesion post abdominal trauma presented a difficult diagnosis, and mucinous appendiceal tumors cannot be excluded despite benign appearance on imaging. A focus on whether the lesion is continuous with the appendix is important, but is not exclusive of appendiceal neoplasms. Pathognomonic signs are present on endoscopy and laboratory tests can be used to supplement diagnosis. Fundamentally, surgical excision is both therapeutic and diagnostic. Surgeons and radiologists alike should both be aware of the pitfalls of imaging when presented with the assessment of a cystic abdominal lesion.

Patient consent

The authors declare that patient informed consent has been obtained in writing for the case report: “Massive Appendiceal Mucinous Neoplasm Diagnosed after Abdominal Trauma: Pitfalls in Imaging and Diagnosis” and is reproducible on reasonable request.

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