Perforated Appendiceal Mucocele within an Amyand’s Hernia: A Case Report and a Brief Review of Literature

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Resumat

Mucocele apendicular perforat într-o hernie Amyand: prezentare de caz și scurtă trecere în revistă a literaturii

Introducere: Hernia Amyand (HA) reprezintă un tip rar de hernie inghinhală, care conține apendicel în sacul herniar, indiferent de prezența apendicitei. Mucocelele apendiculare reprezintă o dilatare a apendicelului cu acumulare de mucus și pot fi un proces fie benign, fie malign. Raportăm un caz excepțional de perforat de mucocele apendicular în cadrul unei HA.

Prezentare de caz: Pacient în vârstă de 77 de ani, a fost supus intervenției chirurgicale pentru o hernie incarcerată. S-a descoperit că avea un apendice dilatat, flegmonos și perforat, dislocat din cec, cu depozite de fibrină și conținut purulent periapendicular în interiorul sacului de hernie. De asemenea, ultima ansă ileală, cecul, colonul ascendent și testiculul drept din sacul de hernie, au fost compromise din cauza procesului septic din jur. A fost efectuată o hemicolectomie dreaptă și orhiectomie dreaptă, urmată de o herniorafie fără însă a se folosi o plasă sintetică. Examenul histopatologic a evidențiat un mucocele apendicular benign perforat asociat cu apendicită flegmonoasă și peritonită fibrinoasă.

Concluzii: Această prezentare de caz descrie o combinație rară de entități: o HA care s-a prezentat ca o hernie incarcerată iar intraoperator a fost evidențiat un apendice dilatat, flegmonos și perforat, care s-a dovedit a fi un mucocele apendicular.

Cuvinte cheie: Hernia Amyand, hernia incarcerată, mucocele apendicular, apendicită flegmonoasă, apendicectomie

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Introduction

Amyand's hernia (AH) is defined as an inguinal hernia, containing the appendix in the hernia sac irrespective of the presence of appendicitis. Appendiceal mucocele represents a dilation of the appendix with an accumulation of mucinous material and could be either a benign or a malignant process. Herein we report an exceptionally rare case of a perforated appendiceal mucocele within an AH.

Case Report

A 77-year-old male patient, underwent surgery for right incarcerated inguinoscrotal hernia. He was found to have a perforated, phlegmonous, and dilated appendix dislocated from the caecum, with peri-appendicular purulent content with fibrin deposits within the hernia sac. Also, the last ileal loop, caecum, ascendant colon, and the right testicle within the hernia sac, were compromised due to the septic process around. A right hemicolectomy and orchiectomy, followed by a herniorrhaphy without using a synthetic mesh, was performed. Histopathological examination revealed a perforated benign appendiceal mucocele associated with phlegmonous appendicitis and fibrinous peritonitis.

Below, we present the case of a perforated appendiceal mucocele associated with phlegmonous appendicitis and fibrinous peritonitis within an AH, following the SCARE criteria (4). Informed signed consent for publication was obtained from the patient.

Key words: Amyand’s hernia, incarcerated hernia, appendiceal mucocele, phlegmonous appendicitis, appendectomy
suggestive of an incarcerated inguinoscrotal hernia. Laboratory workup revealed leukocytosis of 17.2x10^3/μL with neutrophilia. Consequently, the diagnosis of incarcerated right inguinoscrotal hernia with no obstruction was established, with possible vascular damage. As the diagnosis of hernia is purely clinical, no ultrasonography or computed tomography was performed. Thus, the decision was made to proceed to an emergent hernioplasty operation.

The open approach was chosen due to the large hernia dimension. During the surgery, the hernial sac was found, with a thickened and inflamed wall. After opening the sac (Fig. 1), a purulent content with fibrin deposits and a perforated, phlegmonous, and dilated appendix dislocated from the caecum were highlighted.

The terminal ileum loop, entire caecum, ascending colon, and the right testicle were encompassed in the septic process, which prompted their dissection. Consequently, a laparotomy extending medially the initial incision was performed, revealing additional purulent content in the right colic space, with viable colic wall arising only in the transverse colon. In this light, a right hemicolectomy and orchiectomy were performed by the attending surgeon. After lavage of the abdominal cavity and pelvic drainage, the reinforcement of the posterior inguinal wall was made by suturing the conjoint tendon to the inguinal ligament, without using a synthetic mesh due to the septic process.

The patient underwent antibiotic treatment and surgical wound care. The postoperative period was uneventful, and the patient was discharged after 7 days. The patient was followed up for 3 months during which there were no complications. The histopathological exam of the appendix and the hernia sac revealed a perforated benign mucocele associated with phlegmonous appendicitis and fibrinous peritonitis.

**Discussion**

Claudius Amyand was a French-born English surgeon, who described for the first time this particular type of inguinal hernia. On 6 December 1735 at St George’s Hospital, London, he performed the first known appendectomy on an 11-year-old boy, who had an appendicular perforation located within an inguinal hernia sac (1).

Inguinal hernias are one of the most common surgeries that a general surgeon performs. However, AH is a rare type of inguinal hernia, which occurs when the appendix is comprised in the hernia sac. While it is more common in childhood, due to the patency of the processus vaginalis, cases have been recorded in every age group, ranging from 3 weeks to 92 years (5). Most cases are
right-sided, given the typical topography of the appendix (6,7), yet in some cases, such as mobile caecum, a long appendix, malrotation, or situs inversus, the hernia is situated on the left-side (8,9).

The incidence of AH is rare, occurring between 0.19-1.7% of inguinal hernia patients. When it is associated with appendicitis, the rarity becomes even more poignant, with a reported incidence ranging between 0.07-0.13% (10). The particularity of our case consists in reporting not only an extremely rare condition (AH plus appendicitis) but also unique histopathology – a perforated appendiceal mucocele. To the best of our knowledge, no other reports have been published regarding this scenario.

Regarding the pathophysiology of AH, there is debate surrounding the cause of the appendix herniation and subsequently the development of appendicitis within the hernia sac. In a systematic review, the authors suggested that the main cause for the development of appendicitis within an AH is the external compression generated by muscle contraction and the sudden transitory increase in intraabdominal pressure, causing ischemia and the subsequent inflammatory process (11).

Most cases of AH are diagnosed intraoperatively and may be asymptomatic, but some cases may present with incarceration, further complicated by abscess, perforation, epididymitis, or orchitis (12). In our case, a perforated, phlegmonous, and dilated appendix was found in the hernia sac, dislocated from the caecum, with peri-appendicular purulent content and fibrin deposits.

Patients with symptoms and signs of incarceration or strangulation of an inguinal hernia, can be diagnosed by physical exam and rarely require additional imaging. Thus, almost all cases of AH are diagnosed intraoperatively. In a study of 18 consecutive AHs, no patients were diagnosed preoperatively and the most common presenting symptom was inguinal or inguinocrotal swelling (13). However, some studies reported the role of abdominal imaging in the preoperative diagnosis of AH (14). In computed tomography (CT), a tubular blind-ending structure originated from the caecum which extends to the hernia sac, is representative (15,16). In our case, clinical examination revealed a giant right-sided inguinocrotal hernia, therefore we did not additional imaging was deemed necessary.

Different factors establish an appropriate surgical treatment, including appendix condition, characteristics of hernia, and the patient associated diseases. To this point, there is no consensus on whether an appendectomy for a normal appendix should be performed or whether mesh should be used for the hernia if an appendectomy is performed. Depending on the status of the appendix, Losanoff and Basson proposed a classification scheme to determine the management of AH (Table 1) (17).

In most cases, classical treatment is represented by appendectomy via herniotomy with primary hernia repair. When either perforation or pelvic abscess is suspected, lower laparotomy is recommended with or without primary hernia repair (10,17). When a non-inflamed appendix is revealed, most authors recommend appendectomy, while some

<table>
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<tr>
<th>Classification</th>
<th>Description</th>
<th>Surgical management</th>
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<tbody>
<tr>
<td>Type I</td>
<td>Normal appendix in an inguinal hernia</td>
<td>Reduction or appendectomy with mesh hernioplasty</td>
</tr>
<tr>
<td>Type II</td>
<td>Acute appendicitis in an inguinal hernia with no abdominal sepsis</td>
<td>Appendectomy through the hernia, followed by a hernioplasty without a mesh</td>
</tr>
<tr>
<td>Type III</td>
<td>Acute appendicitis in an inguinal hernia and peritonitis</td>
<td>Appendectomy through laparotomy, followed by a hernioplasty without a mesh</td>
</tr>
<tr>
<td>Type IV</td>
<td>Acute appendicitis in an inguinal hernia with concomitant other abdominal pathology</td>
<td>Appendectomy through laparotomy, followed by a hernioplasty without a mesh and other procedures as appropriate</td>
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advocate for prophylactic removal of the appendix due to the risk of re-herniation or future appendicitis (11). There is also a debate regarding mesh placement. The argument against its use following appendicectomy is largely based on the risk of infection, yet if there is no per se appendicitis, there appear to be no counterindications (18,19). Besides the classical approach, there are published studies in which cases of AH have been managed via the laparoscopic approach (20,21). Thus, the literature supports our case-management rationale. We decided against using a mesh, given the extent of the inflammatory process. The laparoscopic approach was not a viable option due to the size of the sac.

Appendiceal mucocele refers to a cystic dilation of the appendix with subsequent accumulation of mucinous material. It can be either a benign or malignant process. Currently, the term “mucocele” might be considered archaic, while the new term of “appendiceal mucinous lesions” is preferred, encompassing two major entity groups: non-neoplastic lesions (mucocele) and neoplastic lesions (22). Optimal management of simple mucocele or mucocele secondary to benign epithelial hyperplasia is straightforward appendectomy (3).

Finally, the classification and treatment of our case did not fit into any one of the Losanoff-Basson types, requiring an outside-the-box approach, tailored to best suit this particular scenario.

Conclusions
The current case report described a trio of rare entities: an AH, complicated with acute appendicitis which proved to be an appendiceal mucocele on the pathology report. The scenario was further complicated by the amplitude of the septic process, which led to an extensive resection and, consequently, to a beyond-available-data approach.

Authors’ Contributions
Călin Crăciun and Flavius Mocian acquired, analyzed and interpreted all the patient data. Călin Crăciun, Flavius Mocian and Marius Coroş contributed to the conception and design of the current manuscript. Rareş Crăciun and Andra Nemeş acquired, analyzed and interpreted the literature references. Flavius Mocian, Rareş Crăciun and Andra Nemeş drafted the manuscript and Călin Crăciun and Marius Coroş revised it critically for important intellectual content. All authors have read and approved the final version to be published.

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Conflicts of Interest
No potential conflict of interest was reported by the authors.

Ethical Statement
Written informed consent was obtained from the patient for the publication of this report.

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