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journal homepage: www.casereports.comAmyand's hernia with acute gangrenous appendicitis and cecal perforation: A case report and review of the literature[☆]William Kromka^{a,*}, Aline S. Rau^b, Charles J. Fox^c^a University of Colorado, School of Medicine, United States^b University of Colorado, United States^c Denver Health and the University of Colorado, United States

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ABSTRACT

INTRODUCTION: An Amyand's hernia is a heterogeneous clinical condition defined by the presence of the vermiform appendix within an inguinal hernia sac, which may or may not contain other abdominal contents or pathologic inflammatory changes. Herein we present an exceptionally rare case of an Amyand's hernia containing acute appendicitis and a perforated cecum.

PRESENTATION OF CASE: A 46-year-old male with a right inguinal hernia of 2–3 year duration presented to our Emergency Department complaining of acute onset abdominal and groin pain. The patient was diagnosed with an incarcerated right inguinal hernia and underwent emergent surgical repair. Intraoperatively a reactive fluid was found within the hernia sac that prompted an exploratory laparotomy for suspected bowel perforation. The hernia was then found to contain an inflamed gangrenous appendix with an inflamed and perforated cecum. An ileocectomy and enteroenterostomy was performed and the hernia defect was repaired without mesh.

DISCUSSION: With an estimated incidence of only 1%, Amyand's hernias are rare and lack a clear evidence-based management scheme. Moreover, they can contain a diverse range of pathologic features and presentations that can complicate diagnosis and treatment. To avoid potential morbidity and mortality, the surgeon must consider an Amyand's hernia on his or her differential when operating on inguinal hernias and be aware of the associated presentations, complications, and management schemes.

CONCLUSION: There is a paucity of reports describing simultaneous appendicitis and cecal perforation within an Amyand's hernia. In our case, ileocectomy and Bassini hernia repair with close follow-up led to a favorable outcome.

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1. Introduction

Groin hernias, particularly inguinal hernias, are one of the most common clinical problems encountered in adults. The estimated lifetime risk of developing a groin hernia is 27% for men and more than 20 million inguinal herniorrhaphies are performed yearly worldwide [1,2]. However, the initial presentation of symptomatic inguinal hernias can masquerade as a variety of underlying disease processes. The case presented here represents the exceptionally rare circumstance of an Amyand's hernia, first described by Claudius Amyand in 1735 as the presence of the vermiform

appendix within an inguinal hernia, with or without appendicitis [3]. This condition has a very low incidence of only 1% and is complicated by acute appendicitis in only 0.08–0.13% of cases [4].

Herein, we report a case of an Amyand's hernia with acute gangrenous appendicitis and a cecal perforation. Also presented is a review and discussion of the literature regarding the pathophysiology, diagnosis, and management of this rare hernia presentation. This work has been reported in line with the SCARE criteria [5].

2. Presentation of case

A 46-year-old male patient presented to the Emergency Department with 24 h of abdominal pain and vomiting with worsening lower abdominal, right groin, and testicular pain and swelling. His past medical history is significant for HIV on HAART with no AIDS defining illnesses and HCV treated with antivirals. He reports a past surgical history of a prior left inguinal herniorrhaphy. The patient reported having known about his present right inguinal hernia for 2–3 years but stated it was only recently that he developed the

Abbreviations: SCARE, surgical case report; HIV, human immunodeficiency virus; HAART, highly active antiretroviral therapy; AIDS, acquired immunodeficiency syndrome; HCV, hepatitis C virus; ED, emergency department; TEP, totally extraperitoneal; CT, computed tomography.

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onset of pain that he described as sharp, constant, and worse with standing or movement.

During examination in the ED, the patient was afebrile and hemodynamically stable and in no apparent distress. His abdominal exam was without rebound or guarding, but the patient did attest to mild tenderness to palpation over the lower abdomen. A right inguinal mass was apparent and the patient had exquisite tenderness to palpation of the right groin and scrotum. The right scrotum was swollen and erythematous. The rest of his physical exam was unremarkable. Laboratory findings included a leukocytosis of 14.4 k/uL, Lactate of 1.6 mmol/L, and a basic metabolic panel within normal limits. A bedside ultrasound was performed in the ED that confirmed an inguinal hernia that contained bowel. Attempts at reduction using 12 mg of morphine were made in the ED without success.

Because the hernia could not be reduced in the ED, the patient was consented for an emergent operative repair of the hernia. After induction of general anesthesia, the fibers of the external oblique were identified and opened, the spermatic cord identified and isolated with a Penrose drain, and the hernia sac identified on the anterior medial border of the spermatic cord without the presence of a direct hernia. The indirect inguinal hernia sac was opened and found to contain incarcerated bowel, omentum, and a malodorous and murky reactive fluid. A subsequent exploratory laparotomy through a midline incision was performed due to a suspected bowel perforation. The small bowel was thoroughly inspected starting from the ligament of Treitz. Ileum was identified entering the internal ring. The hernia was reduced and found to contain an inflamed strangulated appendix with cecal perforation at the origin of the appendix. Thus, an ileocecectomy was performed and a side-to-side functional end-to-end enteroenterostomy was then created with a gastrointestinal stapler. Several liters of warm irrigation were used to lavage the peritoneal cavity. The hernia defect was then repaired without mesh using the Bassini technique. Given the cecal perforation, the abdominal skin incision remained open, the subcutaneous tissue was abundantly irrigated, and a wet-to-dry dressing was then placed between the skin edges.

The patient's postoperative course was uncomplicated. He tolerated advancement of his diet and his pain was well controlled on oral medication. He was discharged to his home on postoperative day five. The patient returned to clinic two weeks following his surgery and was found to be recovering well. Several months later during an outpatient follow up visit, the patient was noted to have a recurrent right inguinal hernia. A successful TEP laparoscopic mesh repair was subsequently performed.

3. Discussion

With an incidence of only 1%, Amyand's hernias are rare occurrences and even less likely to be complicated by acute appendicitis, a finding present in only 0.08–0.13% of cases [4]. One systematic review of the literature indicated that the incidence of Amyand's hernias might actually be smaller than 1%, falling between 0.4% and 0.6%, but the relatively limited number of cases reported makes exact estimates difficult [6]. Due to the relative paucity of cases and a clinical presentation indistinguishable from that of a classic incarcerated hernia, information regarding the pathophysiology, diagnosis, and management of Amyand's hernias is still lacking.

Regarding the pathophysiology of Amyand's hernias, there is debate surrounding the cause of the appendiceal herniation and development of appendicitis within the sac. One explanation for the development of appendicitis within an Amyand's hernia is a previously obstructed appendix, such as from a fecalith or lymphoid hyperplasia, becoming entrapped within a hernia sac and becoming inflamed due to the original obstruction rather than the

entrapment. However, there are only few accounts in the literature of Amyand's appendicitis associated with fecaliths or villous adenoma [7]. Other mechanisms that can lead to acute appendicitis include adhesions causing irreducibility of the hernia and compression of the appendix in the external ring from increases in intra-abdominal pressure [8]. A systematic review of the literature conducted by Michalinos et al. found that most authors believe that the appendicitis develops due to external compression from muscle contraction and sudden increases in intra-abdominal pressure causing ischemia and subsequent inflammation [6].

As in our case presented above, the clinical presentation of Amyand's hernias mimics that of an incarcerated inguinal hernia: a painful irreducible inguinal mass lacking the classic symptoms of acute appendicitis [4]. The absence of signs or symptoms specific to an Amyand's hernia makes clinical diagnosis difficult and leads to a broad differential including strangulated omentocele, Richter's hernia, inguinal adenitis, acute epididymitis, and testicular tumor with hemorrhage. Indeed, Amyand's hernias have been reported to present as blatant surgical emergencies such as a perforated appendix in an inguinal hernia presenting as Fournier's gangrene, but also more subtly such as the giant left-sided Amyand's hernia with an appendiceal abscess without any clinical evidence of incarceration, strangulation, acute scrotum, or occlusion encountered by Mongardini et al. [9,10]. The spectrum of Amyand's presentations is equally diverse in pediatric populations as well, ranging from a neonate reported to have an Amyand's hernia with a perforated appendix and enterocutaneous scrotal fistula to a six-year old boy whose appendix had been perforated by a metal pin [11,12].

Regardless of etiology, these patients with symptoms of an acute incarceration or strangulation of an inguinal hernia can be reliably identified and diagnosed by physical exam and only rarely require imaging studies [1]. Since the standard of care for such patients is emergent surgical repair, it follows that Amyand's hernias are almost exclusively diagnosed intraoperatively. Preoperative diagnosis of an Amyand's hernia is infrequent and has only been reported in a limited amount of cases. For instance, Vehbi et al. used ultrasonography and confirmatory CT scan to successfully diagnose an Amyand's hernia in a 49-year-old female [13] whereas a few others have incidentally, yet successfully identified Amyand's hernias based on preoperative CT scans [14,15].

Once an Amyand's hernia has been identified during surgery, the management can then be guided based on the pathological features present. When there is appendicitis or a perforated appendix, it is widely accepted that an appendectomy should be performed in addition to hernia repair, avoiding the use of mesh due to the danger of infection [6,16]. If a non-inflamed appendix is found, most authors do not suggest removal of the appendix; however, some advocate for a prophylactic appendectomy on the grounds that it is prone to re-herniate and may cause future appendicitis [6]. In order to help guide and improve treatment of Amyand's hernias, Losanoff and Basson devised a widely accepted classification and management scheme that divides Amyand's hernias into four subtypes as demonstrated in Table 1 [17].

Although our patient in the case described above did not have evidence of peritonitis, the murky and malodorous succus encountered in the hernia sac indicated a likely perforation and warranted an exploratory laparotomy for source control. Thus, the classification and treatment of our case failed to categorically fit into one exclusive Losanoff type, yet a favorable patient outcome was still achieved. The Losanoff classification scheme can be a useful tool for initial staging and guidance of management, however our case demonstrates that there is great variability in Amyand's hernia types and treatment needs to be tailored to the particular patient circumstances.

Because Amyand's hernias are typically incidental findings during surgical repair, the surgeon must be prepared to individ-

Table 1
[17].

Hernia Type	Type 1	Type 2	Type 3	Type 4
Clinical Features	Normal appendix	Acute appendicitis within the sac	Acute appendicitis + peritonitis	Acute appendicitis + other abdominal pathology
Surgical Management	Reduction or appendectomy (choose based on patient demographics) + mesh hernioplasty	Appendectomy through hernia, endogenous repair	Appendectomy through laparotomy, endogenous repair	Appendectomy + diagnostic workup for other pathology + additional procedures as needed

ualize treatment accordingly. Indeed, there have been reports of Amyand’s hernias ranging from the entrapment of a simple non-inflamed appendix to a hernia sac containing acute appendicitis with the simultaneous presence of a ruptured cecal adenocarcinoma; the former requiring simple reduction and possible appendectomy depending on surgeon preference and the latter requiring an emergent right hemicolectomy [18]. Though there is a growing collection of literature on Amyand’s hernias, the coexistence of acute appendicitis and a perforated cecum in the hernia sac is not well documented and to the authors’ knowledge this is one of the only instances of such an occurrence followed by an ileocecectomy and Bassini repair that has been reported.

4. Conclusion

Ultimately, the unique variation we encountered underscores the importance of surgical consideration of an Amyand’s hernia during an inguinal hernia repair and individualized treatment guided by the intraoperative findings. As a rare condition with a diverse range of clinical presentations, each reported case of an Amyand’s hernia diagnosis and treatment contributes valuable information that can help improve management and reduce morbidity and mortality through increased physician anticipation and awareness.

Conflicts of interest

The authors of this manuscript have no commercial associations or financial disclosures, such as consultancies, stock ownership, equity interests, patent licensing arrangements, and payments for conducting or publicizing a study that might pose or create a conflict of interest with information presented in this manuscript. None of the authors has a financial interest in any of the products, devices, or drugs mentioned in this manuscript

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Ethical approval

Our institution has exempted ethical approval.

Consent

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.

Author contribution

- William Kromka – Primary author, responsible for literature review and writing the case report.
- Aline Rau – significant editorial contributions
- Charles J. Fox – significant editorial contributions

Guarantor

William Kromka

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