

Amyand's Hernia as a Random Finding in Acute Abdominal Pain and the Role of Thorough Investigation: a Case Report

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ABSTRACT

Objectives: Amyand's hernia is a rare clinical entity, in which the sac of an inguinal hernia contains the appendix. Its incidence is less than 1% of all inguinal hernia cases. Clinical presentation is not specific and can mislead clinical suspicion towards inguinal hernia incarceration. Different classifications have been proposed, with significant importance for the surgical management plan. The main factors to take into consideration include hernia sac content and macroscopic appearance of the appendix, intra-abdominal inflammation, the possibility of hernia recurrence, and patient's general situation and comorbidities.

Materials and methods: A 60-year-old male patient with previous history of tension-free right inguinal hernia repair with mesh and cholelithiasis presented to the Emergency Department complaining of acute abdominal pain localized at the right lumbar and right iliac region and a right groin easily reducible mass. The patient had an abdominal CT performed 48 hours ago that revealed cholelithiasis and a recurrent right inguinal hernia containing the appendix. Because of the consistent pain in the right abdomen despite the easily reducible hernia, a high suspicion of a coexisting pathology was raised and a new abdominal CT scan with intravenous and per os contrast agent was ordered. The latter showed an Amyand's hernia and cholelithiasis, but also detected lithiasis of the right ureter with upstream dilatation as the main cause of abdominal pain.

Results: The patient was subsequently treated with ureteral stent placement and the following day laparoscopic hernia repair with a transabdominal preperitoneal (TAPP) approach and cholecystectomy were performed. The patient was discharged from hospital on the second postoperative day without complications.

Conclusions: Surgeons should be aware of different types of Amyand's hernia and have to include this entity in the differential diagnosis of right iliac fossa abdominal pain combined with a groin mass to avoid intraoperative complications and suboptimal management. It is equally important, though, to exclude

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Article received on the 21st of June 2022 and accepted for publication on the 17th of August 2022

other causes of abdominal pain. This case report reviews a rare entity of right inguinal hernia recurrence presenting as Amyand's hernia during the investigation of right abdominal and iliac fossa pain, that was finally attributed to ureteric colic. Special attention should be given to the thorough investigation of abdominal pain causes.

Keywords: acute abdomen, acute appendicitis, Amyand's hernia, hernia repair, ureterolithiasis, ureteral colic, ureteric stent, case report.

INTRODUCTION

Inguinal hernias are one of the most common surgical entities treated by general surgeons. An Amyand's hernia is an inguinal hernia containing the appendix, with or without inflammation, and has an incidence of 1% of inguinal hernias. Even in the presence of complications, including inflammation, incarceration, perforation, and abscess formation, preoperative diagnosis is challenging (1). It is of critical importance to obtain a preoperative diagnosis, in order to achieve the optimal surgical management plan. The rarity of similar cases and lack of randomized controlled trials render surgical decision making even more demanding.

We present a case of an Amyand's hernia diagnosed preoperatively as a random finding during the investigation of abdominal pain of acute onset due to ureterolithiasis. The patient was managed with placement of a ureteric stent and then laparoscopic transabdominal preperitoneal (TAPP) hernia repair and cholecystectomy. In addition, we present a detailed review of the literature, with emphasis on management regarding Amyand's hernia type.

This case report has been elaborated in accordance with the SCARE criteria for case report publication (2). The patient and his relatives were thoroughly informed and written informed consent was obtained for publication of the present case report and accompanying images. Copies of written consent are available for review by the editor-in-chief of this journal.

CASE PRESENTATION

A 60-year-old male with a history of open right inguinal tension-free hernia repair with mesh seven years ago and cholelithiasis presented to the Emergency Department complaining of acute right iliac fossa pain, which had started

about 72 hours before his arrival, and a right groin mass. The patient denied any other symptoms. He was afebrile, hemodynamically and respiratory stable, with normal total blood count. During clinical examination, an easily reducible recurrent right inguinal hernia and a positive right costovertebral percussion test (Giordano's sign) were noted. Normal bowel sounds were perceptible on auscultation, while digital rectal examination revealed an empty rectal vault, with no evidence of pain. All other hernial orifices were intact. The rest of the physical exam was unremarkable. Patient history and clinical examination led to the suspicion of another coexistent intra-abdominal pathology. A new abdominal CT scan with intravenous and per os contrast agent was performed, which revealed a right Amyand's hernia without signs of inflammation or incarceration (Figure 1), but also indicated lithiasis of the right ureter with upstream dilation (Figure 2). Known medical history of cholelithiasis was also confirmed. Ureter stenting was performed and the following day, after discussing the risks and benefits with the patient, he agreed to proceed with surgery and was taken to the operating room. A laparoscopic transabdominal



FIGURE 1. Abdominal CT showing a right inguinal hernia with a tubular formation inside the hernia sac-Amyand's hernia



FIGURE 2. Abdominal CT showing pyelocalyceal dilation of right kidney due to right ureterolithiasis

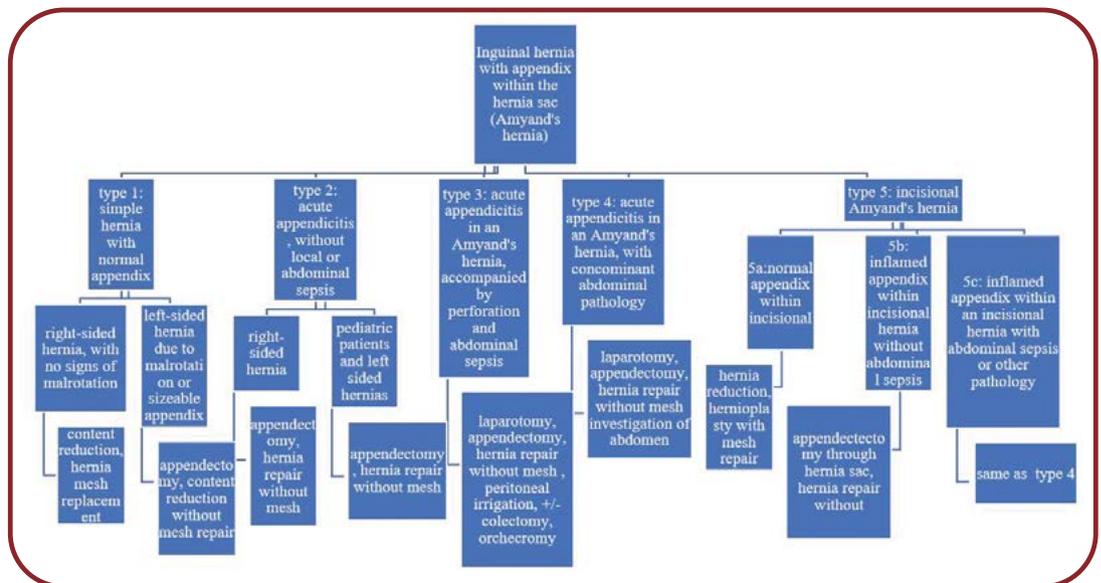
preperitoneal (TAPP) right inguinal hernia repair using a 15 cm x 12 cm polypropylene mesh and cholecystectomy was performed. On gross examination, the appendix was of normal size, without inflammatory or necrotic signs macroscopically. No immediate complications were noted during the postoperative period. The patient was discharged on postoperative day 2. Four years after the operation, the patient is doing well without hernia recurrence.

DISCUSSION

Acute abdominal pain is a frequent symptom among patients presenting to the emergency department. Immediate and careful assessment is necessary to distinguish patients with acute ab-

domen who require urgent surgical intervention from those who must initially be managed conservatively. Besides that, a combination of history, physical examination, imaging and laboratory investigation is necessary to reach the right diagnosis. When a patient's symptoms and condition do not necessitate an urgent operation and when imaging fails to lead to a definitive diagnosis, further abdominal examination by an experienced physician may help to find the underlying pathology (3). The effectiveness of this strategy is obvious in our case. Data from the UK show that patient examination by an experienced surgeon reduces unnecessary hospital admissions (4). Diagnostic laparoscopy can be used in selected cases not only for the diagnosis, but also as a therapeutic approach for various intra-abdominal emergencies such as appendicitis, cholecystitis, adhesiolysis, hernia repair, and many gynecologic emergency conditions (5-7).

The first recorded successful Amyand's hernia management was reported in 1735, being performed by the French surgeon Claudius Amyand at St. George's Hospital in London. Particularly, Dr. Amyand treated a 11-year-old boy who presented with acute inflammation of the vermiform appendix, with the latter being the content of an inguinal hernia (8). Consequently, the pathologic entity of a hernia sac containing the appendix or part of it, whether it is inflamed or not, is named Amyand's hernia. The appendix also has been identified as content of obturator, umbilical and incisional hernia sacs (9). Inguinal



ALGORITHM 1. Algorithm for Amyand's hernia management

hernias constitute one of the most common surgical entities managed by general surgeons, with 700 000 hernia repair procedures annually in the USA (9). Amyand's hernias are very rare in common surgical practice, counting for 1% of all recorded inguinal hernias, with complicated cases such as acute appendicitis, perforation, and necrosis having an incidence of 0.1% (1). In the case of perforated appendix, mortality ranges up to 30% due to severe abdominal sepsis (10). Although Amyand's hernias are diagnosed more commonly during childhood due to patency of processus vaginalis, corresponding in up to 2% of all appendectomies in the neonatal period and childhood, the literature contains reports of Amyand's hernias diagnosed in every age group, from neonates up to elderly people (10, 11). There is a higher incidence among men and it must be highlighted that the literature also contains case reports of Amyand's hernias diagnosed on the left side, which has been attributed to situs inversus, intestinal malrotation, loose cecum or long vermiform appendix (10). Additionally, there are case reports that describe presentation of the appendix in combination usually with the cecum, but also with the bladder, ovaries, fallopian tubes, part of the omentum, or Meckel's diverticulum (10). Furthermore, there are literature reports of appendiceal tumors found within an Amyand's hernia sac (12). Amyand's hernia should not be confused with De Garennot's hernia, which is a femoral hernia containing the appendix (11). Preoperative diagnosis is very challenging and the surgical management plan is usually established intraoperatively, since differential diagnosis is oriented to incarcerated inguinal hernias. Surgeons should be aware of different types of this clinical entity and factors affecting intraoperative decision making such as appendix and hernia macroscopic appearance and characteristics and patient's general situation (1).

According to Losanoff and Basson classification, which was established in 2007, Amyand's hernias can be classified into four discrete types according to the appendix presentation within the hernia sac. Type 1 Amyand's hernia contains a normal appendix within the hernia sac, type 2 hernia is combined with acute appendicitis, but with no signs of perforation, or other source of abdominal sepsis, type 3 hernia is combined with complicated acute appendicitis and intraab-

dominal sepsis, while type 4 Amyand's hernia is accompanied by intraabdominal pathology outside the hernia sac (9).

In 2011, Singal and Gupta proposed a revised version of the above-mentioned classification system, introducing Amyand's hernias that occur in presence of a previous abdominal incision, resulting in this way in type 5, which is an incisional Amyand's hernia. This type was further divided into three subtypes: type 5a, which describes the presence of a macroscopically normal appendix within an incisional hernia sac and is managed similarly to type 1, with simple hernia reduction and mesh repair; type 5b, which corresponds to acute inflammation of an appendix contained in an incisional hernia and should be treated by an appendectomy and primary hernia repair; and type 5c, which represents acute appendicitis within the incisional hernia, with concurrent abdominal sepsis or pathology, that should be treated similarly to type 4 (13). The abovementioned revised classification is also known as Rikki classification (11). Kose *et al* proposed one more type of Amyand's hernia, in which the appendix is connected to the hernia sac with fibrous tissue, but with no signs of inflammation, which also requires appendectomy (1). Taking into consideration the above classifications, our patient presented to the Emergency Department with a type 5 Amyand's hernia.

Amyand's hernias may not present any relevant symptomatology and complications for many years (12). When symptomatic, the clinical presentation of Amyand's hernias is non-specific, mimicking the symptomatology of an inguinal hernia. Usually, a reducible palpable mass of the groin with local pain or discomfort is noticed. When acute appendicitis is present, the patient presents with an irreducible or incarcerated inguinal hernia and symptoms typical of inflammatory process such as nausea, anorexia, vomiting, or right lower quadrant pain. Inflammation can migrate even in the scrotum, with acute symptomatology ipsilaterally with the Amyand's hernia. The clinical examination may reveal, apart from the painful inguinal protrusion, inflammation, and swelling of overlying skin, with absence of McBurney sign.

During the differential diagnosis process, entities that cause painful inguinal swelling, including hydrocele, testicular torsion, inguinal

lymphadenitis, epididymo-orchitis, must be taken into consideration (11).

In case of Amyand's hernia, the pathophysiological mechanisms of inflammatory process and appendicitis include: a) Incarceration and inflammation of the appendix; b) formation of adhesions between the serous membrane of the appendix and the hernia sac, resulting also in incarceration; c) contraction of abdominal wall muscles leads to compression, decreased blood flow and ischemia of the appendix within the sac; d) obstruction of the appendiceal lumen, with consequent edema and inflammation (11); and e) vulnerability to hernia sac trauma and injury (10).

Preoperative diagnosis is extremely challenging, because signs and symptoms can be attributed to incarcerated inguinal hernias, with no specific evidence of appendix participation. In the preoperative setting, trans-abdominal ultrasonography remains a powerful and low-cost diagnostic tool, especially in men with scrotum participation, may reveal a blind ending noncompressible structure with increased vascular flow and wall thickening due to inflammatory process, while the finding of a blind-ending tubular structure that originates from the cecum and ends into the hernia sac, with accompanying wall thickening and periappendiceal fat stranding, in computerized tomography with oral contrast agent may be indicative (9-11). Laboratory tests may reveal inflammatory process or tissue necrosis due to intestinal ischemia (10). It must be noted, although, that CT and US are not part of routine investigation of uncomplicated inguinal hernias preoperatively, thus surgical planning of Amyand's hernias is usually oriented towards the repair of an inguinal or inguinoscrotal hernia (13).

The same classification system is used as a useful guide for management, with a high level of accordance for types 3 and 4, where treatment includes appendectomy combined with hernia repair without the use of mesh (1). Type 1 Amyand's hernia is treated with hernia content reduction and tension-free repair with prosthetic mesh. Apart from lowering the risk of infection, appendectomy avoidance in absence of acute appendicitis may serve for future use of appendix in procedures including extra-hepatic biliary tract replacement, urinary diversion or for Malone procedure, especially in the paediatric

population (10). Left-sided Amyand's hernia type 1 is an exception to this general rule, since a future episode of inflammation may lead to a diagnostic pitfall or even a wrong surgical decision (10). Furthermore, children and adolescent patients may require a prophylactic appendectomy without mesh repair due to a higher life-long incidence of acute appendicitis (2).

Management of types 2, 3 and 4 Amyand's hernias includes appendectomy and primary repair without mesh use, with additional peritoneal irrigation and possible orchiectomy or colectomy for type 3 and pathology investigation and management for type 4 (9). Particularly, performing appendectomy during Amyand's type 2 hernia repair adds a clean-contaminated procedure and therefore increases the risk of infection and inflammation, so mesh use is avoided, with an exception in the treatment of pediatric patients and left side Amyand's hernias. In types 3 and 4 of Amyand's hernias, in which the local environment is complicated by inflammation, pus formation, necrosis, and perforation, prosthetic materials should be avoided in consequence of the threat of surgical site infection and fistulization of the appendiceal stump. Literature references support the performance of the Shouldice technique, because it seems to have lower recurrence rates under these circumstances (10). Another management option for type 3 and 4 hernias is primary appendectomy and primary closure of the inguinal ring, followed by hernia repair with suitable mesh after remission of inflammation (12). Furthermore, the use of minimally invasive techniques promises lower rates of infection and recurrence (11). The evolution of prosthetic materials and biological meshes may provide a viable solution in Amyand's type 2 hernia repair, combining lower infection risk with the elimination of hernia recurrence risk (10).

In addition to the previously mentioned classification, there have been reports for additional types of Amyand's hernias, that pose new challenges to the treatment of type 1 and 2 hernias (1). Kose *et al* present the case of a vermiform appendix incarcerated into the hernia sac without evidence of inflammation that was treated with appendectomy and tension-free hernia repair with mesh use due to low risk of infection and high risk of hernia recurrence. There are also numerous reports of mesh use in Amyand's type 2

hernia repair with no evidence neither of surgical site infection nor hernia recurrence after prolonged follow-up, which may propose a beneficial role of mesh use in type 2 Amyand's hernias, with inflamed but no perforated appendix (1). An overview of surgical management of different Amyand's hernia types is presented in algorithm 1.

Definitive treatment and recurrence-free period depend on the patient's characteristics, initial clinical presentation, and surgeon's perioperative evaluation and experience (5).

More prospective clinical studies about the sensitivity and specificity of CT and ultrasound in the preoperative setting are needed (10). Controversies regarding this rare clinical entity are still present in the surgical community due to the small number of cases reported and lack of randomized controlled studies. Points of controversy include the management of non-inflamed appendix found in an Amyand's hernia and the need of mesh use after appendectomy in type 2 Amyand's hernia (1). □

CONCLUSIONS

Intraoperatively, in cases with inflamed appendix but no signs of perforation, gangrene, or peritonitis, hernia repair with the use of mesh must be taken into consideration by the surgeon, depending on hernia characteristics, patient's general medical state and activity level, and the lifelong risk of hernia recurrence or mesh infection. Ultrasound and CT are useful tools in the armamentarium of the clinician when investigating an inguinal mass preoperatively. The present paper highlights the necessity of bearing Amyand's hernia in mind when managing inguinal hernias, especially right-sided ones, but it also highlights the necessity of investigating other sources of abdominal pain. □

Conflicts of interest: none declared.

Financial support: none declared..

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