Amyand's hernia complicated with acute appendicitis:
A case report and literature review

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Abstract
Amyand's hernia is the presence of the appendix within an inguinal hernia sac. It is a rare condition, occurring in 1% of inguinal hernia patients. The clinical presentation varies depending on the extent of appendix inflammation. Amyand's hernia is difficult to diagnose clinically. However, imaging studies are valuable for both its diagnosis and detection of the associated complications. Here, we report a case of Amyand's hernia in a 3-year-old male child who presented with a history of right inguinal tenderness, pain, and swelling. An operation revealed a hernia sac containing the inflamed appendix; hence, an appendectomy was performed along with a right inguinal herniotomy.

Key Words
Inguinal hernia; Amyand’s hernia; acute appendicitis.

INTRODUCTION
Amyand’s hernia is the presence of the vermiform appendix within an inguinal hernia sac; this condition was first described by Claudius Amyand in 1736 [1,2]. In the literature, these types of hernias have a reported incidence of approximately 1% of all hernias, and these are complicated by acute appendicitis in 0.08% of the cases [3-5]. An Amyand’s hernia can be found with an inflamed, infected, or perforated
appendix. The clinical findings of an Amyand’s hernia usually mimic an irreducible or strangulated hernia and therefore, the vast majority of patients are not diagnosed preoperatively [6]. Hence, these cases are most commonly detected incidentally during a hernia repair. Imaging is also valuable in the diagnosis and detection of the associated complications [7]. Ultrasonography (USG) may be useful if the appendix is suspected to be in the sac and/or the hernia is irreducible [8]. Here we present a rare case of Amyand’s hernia complicated with acute appendicitis in a 3-year-old male child who presented with pain, tenderness, and right inguinal swelling.

CASE REPORT

A 3-year-old male child presented to our emergency department with a 2-day history of pain and swelling in the right groin. Physical examinations revealed a swelling with pain and tenderness in the right groin, and an irreducible right inguinal hernia. There were no bowel symptoms. However, on abdominal examination, there was tenderness in the right lower quadrant. He had no fever, and his white blood count was 11,000/mm³. A direct X-ray of the abdomen was unremarkable. Urgent ultrasonography of the right groin showed a blind ending tubular structure in the right groin sac. The patient was operated on for an irreducible hernia and perioperatively, the inflamed appendix was found within the hernia sac (Fig. 1).

Fig. 1. The operation showed inflamed appendix within the right-sided hernia sac.

A classic appendectomy and herniotomy was performed. The patient was discharged from the hospital without complications on the second postoperative day. In a follow-up one week later, the right inguinal region appeared to be healing well. The histopathology examination reported an acute appendicitis.

DISCUSSION

Amyand’s hernia is an extremely rare condition and has been reported in patients ranging in age from 3 weeks to 92 years. The appendix is found in the hernial sac in about 1% of inguinal hernias and an
inflamed appendix is found in only 0.13% of cases. Claudius Amyand (1681–1740) was the first to perform an appendectomy [5]. Additionally, Amyand’s hernia in children less than 1 year old represents 2% of the total cases of appendicitis [9]. Most cases of Amyand’s hernia occur on right side because of the normal anatomical position. Left-sided Amyand’s hernia is usually associated with congenital anomalies like situs inversus and malrotation. Losanoff and Basson [10] proposed a classification scale to identify and treat Amyand’s hernias. Type I is a normal appendix in which a reduction or appendectomy with mesh hernioplasty should be performed. Type II is an acute appendicitis localized in a hernial sac, which is treated with an appendectomy through the hernia with a mesh hernia repair; however, this procedure is associated with a higher risk of mesh infection. Type III is an acute appendicitis complicated by peritonitis, which is repaired with an appendectomy through laparotomy; the hernioplasty decision in these cases should be made based upon the spread of sepsis. Type IV is an acute appendicitis accompanied by another abnormal pathology; in these cases a hernioplasty may be contraindicated if the damage is too extensive. Our case was type II according to the Losanoff and Basson classifications.

The pathophysiology of Amyand’s hernia and its relationship with appendicitis are unknown [9]. However, many authors believe there is an association between incarceration of the appendix in the inguinal canal and inflammation of the cecal appendix in the hernial sac [11,12]. It is thought that the appendix of Amyand’s hernia is more likely to become inflamed, especially when compared to an anatomically normal appendix since it is retained in that location by adhesions and becomes vulnerable to trauma [13]. It has been suggested that appendiceal migration into the inguinal canal may increase vulnerability to trauma and a compromised blood supply, followed by generalized inflammation and bacterial overgrowth [14]. Additionally, contraction of the abdominal muscles and other sudden increases in intra-abdominal pressure may cause compression of the appendix, resulting in further inflammation [15].

The clinical presentation of Amyand’s hernia is variable and is dependent on the degree of inflammation and whether it presents with or without a perforation. Hence, it may be found with a clinical picture as being normal, obstructed, or strangulated inguinal hernia and an acute scrotum. Common complaints include sudden-onset of epigastric or periumbilical
pain with localized tenderness in the right lower quadrant, combined with a tender irreducible mass in the inguinal or inguinoscrotal region [16]. Additionally, since there are no specific findings with Amyand’s hernia, it may mimic a strangulated inguinal hernia, testicular torsion, epididymo-orchitis, and inguinal lymphadenitis.

In some cases, abdominal exams, physical signs, lab results, and imaging have proven helpful in differential diagnosis. Preoperative diagnosis of Amyand’s hernia is rare, and diagnosis is generally made during a surgical exploration of the groin [9,17]. In our previous study on Amyand's hernia, on 12 (57.1 %) patients who had received USG, Amyand’s hernias were diagnosed in nine patients using preoperative inguinal USG that revealed a blind ending intestinal loop, similar to the present case [8]. USG may be useful in cases in which there is a blind ending tubular structure in the inguinal canal on physical examination, and this application can increase the frequency of radiologic diagnosis of Amyand’s hernia [8,17]. Although a preoperative CT of the abdomen may be helpful in securing the correct diagnosis, this is not a routine practice after the clinical diagnosis of a complicated hernia [13]. Surgical modalities for Amyand’s hernia depend on the inflammatory state of the appendix. While some studies believe in the necessity of an appendectomy even when the appendix is not inflamed; this is mainly meant to avoid future complications [18]. Others advocate for an appendectomy only if the appendix is inflamed [17,19,20]. Because, the appendix lately could be used for biliary tract reconstruction, urinary diversion, anterograde bowel enemas, etc. [8, 21]. Manipulation of a normal appendix during surgery may induce inflammation and may stimulate secondary appendicitis [18,22]. However, at least one study has suggested that laparoscopic procedures involving the manipulation of the appendix do not increase the incidence of appendicitis [16]. In conclusion, Amyand’s hernia is rare and remains a surgical enigma. Here, we report a very rare case of a 3-year-old male with Amyand’s hernia who presented with swelling in the right groin, tenderness, and pain. This condition is difficult to diagnose clinically, and diagnosis can be aided by a high index of suspicion. USG may be a valuable tool in the preoperative diagnosis of Amyand's hernia. In cases of Amyand's hernia complicated by acute appendicitis, it necessitates an appendectomy via the hernial sac.
Acknowledgements
The author(s) declare that they have no competing interests and financial support.

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