A clinical case of successful treatment of acute phlegmonous appendicitis incarcerated in a hernial sac (Amyand’s hernia)

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Atypical localization of the vermiform appendix (VA) in acute appendicitis can lead to diagnostic and tactical errors in emergency abdominal surgery. The rarest and most atypical localization of the VA is within the hernial sac. This type of hernia is defined as Amyand’s hernia. The optimal scope of surgical intervention and access method (laparoscopic or open) for Amyand’s hernia have not been definitively determined. Questions remain regarding the necessity of removing an unaffected vermiform appendix, the approach to hernioplasty in destructive appendicitis, and the feasibility of using a mesh implant for hernioplasty. In cases of a sliding hernia, the destruction of the hernial sac can lead to difficulties with suturing the peritoneum, assessing the state of the strangulated testicle in men, and performing an orchidectomy. Therefore, it is imperative to establish an algorithm for the evaluation and surgical management of patients with strangulated inguinal hernias.

Objective — present a clinical case of successful treatment of Amyand’s hernia with acute phlegmonous appendicitis incarcerated in a strangulated inguinal hernial sac and reactive orchoepididymitis, focusing on the selection of medical and diagnostic strategies to reduce the risk of complications.

The clinical case illustrates the effective management of a patient who presented with acute phlegmonous appendicitis and purulent orchoepididymitis. These abnormalities were found to be atypically localized within the sac of a strangulated right-sided inguinal hernia, known as Amyand’s hernia. The necessary surgical procedures included an appendectomy and an orchidectomy.

Conclusions. Inflammation of the vermiform appendix in Amyand’s hernia can lead to various unpredictable purulent complications, such as acute typhlitis, purulent peritonitis, necrotizing orchoepididymitis, and Fournier’s phlegmon, necessitating extensive surgical intervention. The collective global and personal experience in treating Amyand’s hernia suggests the possibility of establishing standardised protocols for diagnosing and selecting a surgical intervention technique.

Keywords
inguinal hernia, acute appendicitis, Amyand’s hernia, complications, orchoepididymitis.

Atypical localization of the appendiceal appendix within or outside the abdominal cavity can cause diagnostic and tactical errors in emergency abdominal surgery. Examples of such localizations are the presence of the appendix in the hernial sac of a right-sided inguinal hernia (Amyand’s hernia) and a right-sided femoral hernia (De Garengeot’s hernia) [5–7]. A rare occurrence is the presence of the appendix inside an inguinal hernia, referred to as Amyand’s hernia, with a prevalence ranging from 0.07 % to 4 % [2, 9, 11, 13, 15, 19]. S. Amyand, an English military surgeon, was the first to describe such a case. In 1735, an 11-year-old child underwent surgery at St. George’s Hospital in London for a severe case of appendicitis that was complicated by a faecal fistula. The appendix was found in an inguinal hernia [8, 10, 12].

The reasons that usually lead to the specified localization of the appendix are not exactly known, but it is believed that the risk factors may include:

1. Gender: men suffer from this pathology more often.
2. Age (the elderly and children), which is explained by the increased mobility of the intestine and atrophy of the tissues of the posterior wall of the inguinal canal [4, 32].

3. Associated diseases: diseases of the respiratory system, obesity, constipation.


5. Long-term presence of an inguinal hernia.

The age of patients varies from 3 weeks to 92 years, as reported in the literature. The incidence of Amyand’s hernia with concurrent acute appendicitis ranges from 0.07 % to 0.13 % [1, 17]. In this situation, the symptoms of acute appendicitis are prevalent. Mortality rates range from 5 % to 30 % and are influenced by the occurrence of complications resulting from acute appendicitis, such as severe peritonitis, an intestinal fistula, or Fournier’s phlegmon [23, 25].

However, the location of the appendix in the hernial sac does not always lead to the development of acute appendicitis [14, 16, 18].

**Objective** — present a clinical case of successful treatment of Amyand’s hernia with acute phlegmonous appendicitis incarcerated in a strangulated inguinal hernial sac and reactive orchoepididymitis, focusing on the selection of medical and diagnostic strategies to reduce the risk of complications.

**Clinical case**

Patient A., born in 1939, with case history No. 3573, was urgently admitted to the surgical department of the Kyiv Road Clinical Hospital No. 1. Upon admission, the diagnosis was a «strangulated right-sided inguinal hernia». Complaints of a painful lump in the right inguinal area that developed following physical exertion three days prior to hospitalisation. The anamnesis reveals that the patient has had a right-sided inguinal hernia for the past 15 years. On examination: the patient’s general condition is relatively satisfactory; the skin and visible mucous membranes are normal in colour. Hemodynamics is stable. Respiratory rate: 18/min. Blood pressure: 130/70 mm Hg. Above the lungs, breathing is vesicular. The abdomen, which is symmetrical, participates in the act of breathing. Pain in the right inguinal area, where a dense painful mass measuring 10 × 6 cm is palpable and does not extend into the abdominal cavity; the cough symptom is negative. Auscultation: bowel sounds. The symptoms of peritoneal irritation are negative. Diuresis and physiological discharge are within normal limits. The patient was examined clinically, laboratory, and radiologically. The diagnosis was established: a strangulated right-sided inguinal hernia.

Urgent surgical intervention under spinal anaesthesia was suggested and performed. The surgeon accessed the right inguinal area and isolated the seminal cord in layers, discovering a tense hernial sac of 12 × 9 × 7 cm. Its contents included the dome of the cecum with a phlegmonous appendix (Fig. 1). The tip of the appendix was located within a periappendicular abscess, known as Amyand’s hernia. A typical appendectomy was conducted. An additional examination of the hernial sac contents revealed reactive purulent orchoepididymitis, necessitating a right-sided orchidectomy (Fig. 2).
Basini’s hernioplasty was performed on the right with layer-by-layer suturing of the wound. The postoperative period was uneventful. On the 7th day, the patient was discharged in satisfactory condition and received outpatient care.

Pathological and histological examination: phlegmonous appendicitis. Areas of fibrous and fatty tissue with foci of purulent inflammation and structureless masses with the accumulation of segmented leukocytes. Purulent-necrotic orchoepididymitis.

Discussion

The pathophysiology of acute appendicitis within Amyand’s hernia is still controversial [30]. External obstruction, rather than internal obturation of the appendiceal lumen at the neck of the hernial sac, is commonly regarded as the primary cause of acute appendicitis [22, 25]. According to Abu-Dalu and Urcu, appendix trauma is caused by abdominal muscle contraction and increased intra-abdominal pressure, which disrupts blood supply, resulting in tissue ischemia and the development of acute inflammation in the appendix [21, 22, 31]. In 2008, J. Losannoff and M. Basson created a morphological classification of Amyand’s hernias, identifying four potential combinations of an inguinal hernia and an appendix [20, 27, 29, 30] (Table 1).

Later, the classification was supplemented by the inclusion of cases involving the location of the appendix in a postoperative hernia [28] (Table 2). Despite existing classifications, the issue of surgical strategies for these patients remains debatable. Is it necessary to remove an unaffected appendix from the hernial sac? What are the hernioplasty techniques in destructive appendicitis? [3, 34].

We support the idea of other authors [3, 14, 20, 28] that in the presence of purulent inflammation,

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**Table 1. Amyand’s classification of hernias [20]**

<table>
<thead>
<tr>
<th>Classification</th>
<th>Description</th>
<th>Surgical management</th>
</tr>
</thead>
<tbody>
<tr>
<td>Type 1</td>
<td>Normal appendix within the hernial sac</td>
<td>Hernioplasty with a possible situational appendectomy, depending on the age of the patient</td>
</tr>
<tr>
<td>Type 2</td>
<td>Acute appendicitis within an inguinal hernia, no peritonitis</td>
<td>Appendectomy through hernioplasty, hernioplasty with own tissues of the hernial gate</td>
</tr>
<tr>
<td>Type 3</td>
<td>Acute appendicitis within an inguinal hernia, the presence of peritonitis</td>
<td>Appendectomy through laparotomy, hernioplasty with own tissues of the hernia</td>
</tr>
<tr>
<td>Type 4</td>
<td>Acute appendicitis within an inguinal hernia, the presence of other abdominal diseases</td>
<td>Treatment tactics are the same as for types 1–3, and appropriate examination and treatment of other diseases of the abdominal cavity</td>
</tr>
</tbody>
</table>

**Table 2. Expanded classification of Amyand's hernias [28]**

<table>
<thead>
<tr>
<th>Classification</th>
<th>Description</th>
<th>Surgical management</th>
</tr>
</thead>
<tbody>
<tr>
<td>Type 1</td>
<td>Normal appendix within an inguinal hernia</td>
<td>Hernioplasty, appendicectomy in young patients</td>
</tr>
<tr>
<td>Type 2</td>
<td>Acute appendicitis within an inguinal hernia, no abdominal sepsis</td>
<td>Appendicectomy through hernia, primary repair of hernia, no mesh</td>
</tr>
<tr>
<td>Type 3</td>
<td>Acute appendicitis within an inguinal hernia, abdominal wall, or peritoneal sepsis</td>
<td>Laparotomy, appendicectomy, primary repair of hernia, no mesh</td>
</tr>
<tr>
<td>Type 4</td>
<td>Acute appendicitis within an inguinal hernia, related or unrelated abdominal pathology</td>
<td>Manage as types 1 to 3 hernia, investigate or treat secondary pathology as appropriate</td>
</tr>
<tr>
<td>Type 5a</td>
<td>Normal appendix within an incisional hernia</td>
<td>Appendicectomy through hernia, primary repair of hernia including mesh</td>
</tr>
<tr>
<td>Type 5b</td>
<td>Acute appendicitis within an incisional hernia, no abdominal sepsis</td>
<td>Appendicectomy through hernia, primary repair of hernia</td>
</tr>
<tr>
<td>Type 5c</td>
<td>Acute appendicitis within an incisional hernia, abdominal wall, or peritoneal sepsis or in relation to previous surgery</td>
<td>Manage as type 4</td>
</tr>
</tbody>
</table>
it is appropriate to use autoplasty, which was performed in this study.

Amyand’s hernia is a rare disease that is difficult to diagnose in our preoperative stage. In our study, it was not diagnosed before surgery. There are also reports in the literature about the left-sided location of Amyand’s hernia [24]. The primary cause of Amyand’s hernia has not yet been clarified. The presence of acute appendicitis in the hernial sac increases the risk of developing complications. In our case, the patient had phlegmon of the hernial sac and acute orchephepidymitis. The issue of the scope of surgical intervention is debatable. No definitive methodology has been established for examining patients using additional imaging techniques such as ultrasonography and computed tomography before surgery, which could help determine the surgical approach and scope of intervention.

Conclusions
Inflammation of the vermiform appendix in Amyand’s hernia can lead to various unpredictable purulent complications, such as acute typhlitis, purulent peritonitis, necrotizing orchephepidymitis, and Fournier’s phlegmon, necessitating extensive surgical intervention.

The collective global and personal experience in treating Amyand’s hernia suggests the possibility of establishing standardised protocols for diagnosing and selecting a surgical intervention technique.

DECLARATION OF INTERESTS
The authors declare that they have no conflicts of interest.

AUTHORS CONTRIBUTIONS
Conception and design, drafting the article: O. I. Haluzynska; acquisition of data, analysis and interpretation of data, conception and design, drafting the article: O. I. Haluzynska; acquisition of data, analysis and interpretation of data: E. V. Lychak, O. I. Haluzynska; conception and design, drafting the article: O. I. Haluzynska; acquisition of data, analysis and interpretation of data: L. S. Bilianskyi; writing—original draft: O. I. Haluzynska; critical revision of the article: О. І. Haluzynska, L. S. Bilianskyi; writing—original draft: O. I. Haluzynska; critical revision of the article: О. І. Haluzynska, L. S. Bilianskyi; acquisition of data, analysis and interpretation of data: Сonception and design, drafting the article: О. І. Haluzynska.

REFERENCES
Клінічний випадок успішного лікування гострого флегмонозного апендициту, локалізованого в грижовому мішку (грижа Аміанда)

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Нетипова локалізація червоподібного відростка (ЧВ) у разі гострого апендициту може спричиняти діагностичні й тактичні помилки в ургентній абдомінальній хірургії. Найрідкіснішою та нетиповою локалізацією ЧВ є його розташування в грижовому мішку — hernia of Amyand. Досі не з'ясовано, яким має бути обсяг і сутність операції в разі hernia of Amyand: чи необхідно видаляти незмінений ЧВ, як виконати герніопластику при деструктивному апендициті, чи можливе використання сітчастого імплантата для герніопластики. Враховуючи деструкцію грижового мішка при ковзній грижі можуть виникнути складності з ушкодженням яєчка, оцінюванням стану защемленого яєчка у чоловіків, необхідністю виконання орхідектомії. Таким чином, окреслюється необхідність визначення алгоритму обстеження та хірургічного лікування пацієнтів з защемленими паховими грижами.

Мета — на прикладі ілюстрації клінічного випадку успішного лікування грижи Аміанда з флегмонозно зміненим ЧВ у вмісті защемленої пахової грижі і реактивним орхепідидимітом привернути увагу до вибору лікувально-діагностичної тактики, що сприятиме зниженню ризику ускладнень.

Клінічний випадок демонструє досвід успішного лікування хворого з гострим флегмонозним апендицитом та грижою Аміанда. Об’єм хірургічного втручання вимагав виконання апендектомії та орхідектомії.

Висновки. Гостре запалення ЧВ при грижі Аміанда може сприяти розвитку непередбачуваних гнійних ускладнень, а саме, гострого тифліту, гнійного перитоніту, некротичного орхепідидиміту та флегмони Фурньє, що вимагає виконання непередбачуваного об’ємного хірургічного втручання. Накопичений в світі та власний досвід лікування грижі Аміанда вказує на необхідність стандартизації підходів до діагностики та вибору методу хірургічного втручання.

Ключові слова: пахова грижа, гострий апендицит, грижа Аміанда, ускладнення, орхепідидиміт.