



## Amyand Hernia Found in an Infant, Case Report and Review of the Literature

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### Abstract

Amyand's hernia refers to the presence of the appendix inside an inguinal hernia. It is mainly a trans-operative find, affecting less than 1% of inguinal hernias. We report a 4-month-old infant, initially presenting an irreducible right-sided inguinal hernia. Due to the incarceration, surgery was performed. The finding occurred incidentally during the procedure; a conservative approach was preferred, preserving the appendix and returning it to the abdominal cavity, before repairing the inguinal defect. The patient continued asymptomatic at the 12-month follow-up. Despite its low incidence, this condition should be considered when dealing with an irreducible right-sided hernia, especially in infants.

**Keywords:** Appendix; Hernia; Infant; Amyand hernia

### Introduction

First described by Claudius Amyand in London, 1735, while operating on an eleven-year-old boy, Amyand's hernia is a rare disorder in which the appendix can be found inside an inguinal hernia [1,2]. Its current incidence is estimated to range between 0.19% and 1.7% of all cases of inguinal hernia, all the while being up to 3 times more frequent amongst infants, and even more rare when an inflamed appendix is involved [3,4]. It is usually described as a surgical finding due to its non-specific presentation, therefore requiring a high degree of suspicion for a preoperative diagnosis. The clinical picture most frequently encountered is very similar to that of an incarcerated right-sided inguinal hernia, with an irreducible mass that may or may not be tender to touch [3-5]. Laboratory studies usually show either no signs of systemic inflammation or an increase in inflammatory cell numbers, with a predominance of neutrophils [6]. Computed tomography is the current diagnostic gold standard in cases where the diagnosis is considered before entering the operating room [7]. Due to its extremely low frequency, a general consensus on its management isn't currently defined and several schools of thought exist regarding proper treatment, without significant randomized clinical trials existing to support each recommendation. Here, we report the case of a 4-month-old infant who underwent a routine inguinal hernia repair, with the consequential finding of the vermiform appendix inside of the hernia sac.

### Case Presentation

A healthy 4-month-old male infant was brought by his mother, who noticed a bulge in his right scrotum two weeks before arriving at the hospital. Upon examination, the bulge is irreducible, does not transilluminate and is slightly tender to touch. An incarcerated hernia is suspected, and the infant is programmed for surgery. Blood studies do not show a rise in white cells or any other signs of inflammatory response. No imaging studies are ordered. An oblique, 2.5 cm incision is made along the right inguinal region, dissecting the subcutaneous tissue until reaching the aponeurosis of the external oblique muscle, once there, a 0.5 cm hernia sac is found. An initial attempt of manually reducing the defect fails, enticing the dissection of the sac, inside of which a 2.5 cm × 0.5 cm appendix is found (Figure 1). Upon visual examination, no signs of inflammation, abscess, necrosis or perforation are found, therefore, the appendix is returned to the abdominal cavity and the defect is repaired with the Shouldice technique. The patient is monitored over the course of the following two days, showing an absence of inflammatory signs, adequate oral intake and appropriate passing of stool, being discharged on his third day of hospital stay. No recurrence is recorded after 12

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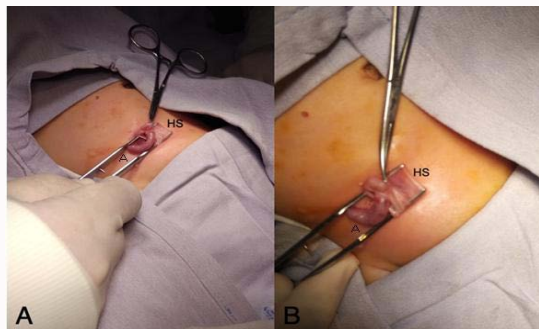
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**Figure 1:** A) Trans-surgical exploration of the hernia sac reveals the presence of a 2.5 cm × 0.5 cm appendix inside of it, without signs of inflammation or perforation. It is diagnosed as a type 1 Amyand hernia, opting to return the appendix to the abdominal cavity without excision. Afterwards, the inguinal defect was repaired without the use of a prosthetic mesh due. B) Closer view of the vermiform appendix protruding from within the hernia sac during its dissection. (A: Appendix; HS: Hernia Sac).

Type	Definition	Management options
1 <sup>a</sup> /A <sup>^</sup>	Non-inflamed appendix inside an inguinal hernia	Hernioplasty with mesh repair, without appendectomy
2 <sup>a</sup> /B <sup>^</sup>	Acute appendicitis inside hernia sac, without abdominal sepsis	Appendectomy plus hernioplasty without mesh repair
3 <sup>a</sup> /C <sup>^</sup>	Acute appendicitis inside hernia sac, with either an abdominal wall or peritoneal sepsis	Laparotomy, appendectomy, primary repair without mesh
4	Acute appendicitis inside hernia sac, with concurrent abdominal pathology	Manage concurrent pathology according to diagnosis + Type 2-3 management
*Atypical presentation	Left Amyand's hernia (situs inversus, intestinal malrotation, mobile cecum with long mesentery and an appendix that crosses the midline)	Consider appendectomy to avoid future diagnostic issues

**Figure 2:** Classifications for Amyand's hernia and current management options according to the literature.

<sup>a</sup>Types from 1 to 4 (Losanoff-Basson)

<sup>^</sup>Types from A to C (Fernando and Ceulemans)

\*Atypical presentations of Amyand's hernia are not included in either classification but are mentioned for the sake of including all current diagnostic possibilities and management options

months of follow-up (Figure 2).

## Discussion

Out of the 20 million hernia repairs performed yearly, an estimated 1% may present with the appendix inside of the hernia sac [5]. It can affect patients of all ages, with some series indicating a predominance of older patients, but most studies suggesting a higher rate amongst infants, with a mean age of 16 months, mainly attributed to a patent vaginal process or another type of congenital anomaly. Much like in inguinal hernia, where the rate of affection is significantly higher amongst men, with an up to 9:1 male to female ratio, Amyand's hernia seems to also affect males much more predominantly [8-10]. Despite this, a proper ratio is yet to be determined. Diagnosis is usually made during the surgical procedure, with the rates of preoperative diagnosis being as scarce as 0.16% in a study that spanned 40 years of case-recording. Abdominal ultrasound can be a safe, fast diagnostic method which could allow the visualization of a cylindrical shape inside the hernial sac. A higher sensitivity is achieved via the use of computed tomography, at the cost of exposing the patient to high-dose radiation; typical images will show the appendix inside the hernia sac, or the ileocecal valve as being closely attached to the inguinal region. Laboratory studies usually do not correlate to actual inflammatory status due to the hernia sac acting as insulation against systemic inflammation. A classification was proposed by Losanoff and

Basson, which uses the appendix status as a guideline for establishing the best treatment option. Type 1 encompasses a normal appendix inside an inguinal hernia, type 2 is equivalent to an acute appendicitis inside the sac without abdominal sepsis, and type 3 involves acute appendicitis and an abdominal wall or peritoneal sepsis, with type 4 presenting both an acute appendicitis and an accompanying abdominal pathology. The appropriate surgical management for each type has been a point of debate in recent years. The majority of opinions agree in the preservation of the appendix in most cases presenting with a type 1 hernia. Notable exceptions for this rule could be anatomical variants in patients which could lead to a delayed diagnosis of appendicitis further on in life [9]. The main reason cited to preserve the appendix is the avoidance of turning an otherwise class 1 or *clean* procedure into a class 2 or *clean-contaminated* one, which in turn translates into an increased risk of infection, fistulization and/or hernia recurrence, while also allowing for the use of a prosthetic or biological mesh during hernia repair [10]. Type 1 and 2 may be treated as minimally invasive or open surgery, while type 3 strictly requires open surgery and type 4 is left to the surgeon's choice, due to its variability in etiology. Laparoscopic approach should be avoided in cases presenting with intestinal obstruction or type 2 scenarios with a high suspicion of perforation [11]. A similar classification was proposed by Fernando and Ceulemans, which encompasses the first three types of the Losanoff and Basson classification, with a similar surgical approach, the main difference being the exclusion of type 4 hernia [12]. Prognosis is dependent on the degree of appendix affection, with hernia recurrence and infection rates being low for type 1 cases, and type 2-3 reporting rates of surgical site infection of up to 9% during community surveillance. Type 4 prognosis depends on the concomitant pathology and patient factors, therefore it must be properly analyzed and discussed with each patient [12,13].

## Conclusion

In conclusion, while a rare presentation, Amyand's hernia must be taken into consideration when approaching the differential diagnosis of a non-reducible right-sided inguinal hernia. The optimal treatment for each case must be individualized according to the clinical context of each patient, with the inflammatory state of the appendix during surgery being the main point factored into the decision. In regard to the management of the appendix, the authors of this paper suggest reserving appendectomy for types 2 to 4 and type 1 hernias with an unusual presentation, such as those left-sided. As far as the use of a prosthetic mesh is concerned, it should be reserved for cases where the wound is either kept clean, proper antibiotic coverage is employed or patient benefit outweighs infection risks, such as high risk of hernia recurrence.

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