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Amyand's Hernia in Children: Two Rare Cases and Review of the Literature

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Abstract **Case Report**

Amyand's hernia is rare in children. Acute appendicitis associated with this hernia is even rarer, occurring in only 0.08% of cases [3]. Clinical symptoms may vary and often resemble those of an incarcerated inguinal hernia, which may lead to a misdiagnosis prior to surgery. We report 2 cases of Amyand's claudius hernia operated on at the Souss Massa Agadir university hospital, Morocco. The first case was a 3-year-old boy scheduled for surgical cure of a simple right inguino-scrotal hernia, where a non-inflammatory appendix was revealed within the hernia sac. The second case was a 22-day-old newborn admitted to the emergency department with a strangulated right inguino-scrotal hernia. The diagnosis of Amyand's hernia was made only after surgical exploration, during which the pathological appendix was discovered in the hernia sac. This underlines the importance of considering this rare pathology when evaluating inguinal hernias, particularly in children.

Keywords: Amyand's hernia, acute appendicitis, inguinal hernia, child.

INTRODUCTION

Amyand's hernia is the presence of a vermiform appendix inside an inguinal hernia. This pathology was first described by Claudius Amyand in 1735 at Saint George's Hospital in London, where it was observed in an 11-year-old child, admitted with a right inguinal hernia complicated by a right stercoral fistula [1].

The incidence of this pathology is only 1% [2], and acute appendicitis complicates it in only 0.08% of cases [3]. Intraoperative diagnosis is rare, as it is an incidental finding [4]. It is often diagnosed clinically as a strangulated hernia [5].

We report the first 2 cases of Amyand's hernia at the University Hospital of Souss Massa Agadir, the diagnosis of Amyand's hernia was made intraoperatively in both cases, hence the importance of considering Amyand's hernia as a possible diagnosis for inguinal hernias, particularly in children.

Case 1:

A 3-year-old boy, with no notable pathological history, presented to the pediatric surgery department of University Hospital of Souss Massa Agadir with a right inguino-scrotal tumefaction, painless, expansive on crying, reducible on palpation and non-inflammatory in appearance, evolving for 1 year, with no other associated digestive signs. Abdominal examination revealed a soft abdomen. Examination of the external genitalia showed that the testicles were in place and the transillumination test was negative. The presumptive diagnosis was an uncomplicated right inguino-scrotal hernia.

In dorsal decubitus under general anaesthesia, an skin incision was made in the right lower abdominal fold, and exploration revealed a vermiform appendix adhering to the wall of the hernia sac. We performed an appendectomy after meticulous dissection of the hernia sac wall to isolate the appendix (Figure 1), followed by ligation of the hernia sac.

The post-operative course was straightforward, and the child was discharged after 48 hours in hospital.

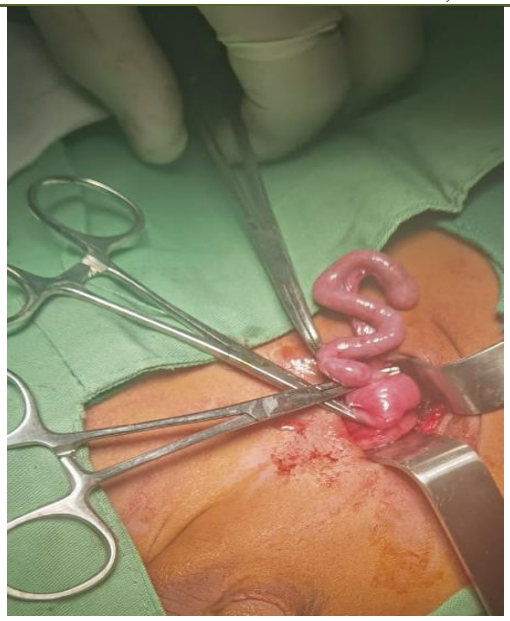

Figure 1: Intraoperative image showing inflamed appendix in the right hernia sac

Case 2:

The patient was a 22-day-old male newborn with no notable pathological history, admitted with a 2-day history of febrile occlusive syndrome.

On admission, the newborn was conscious, pink, hypotonic, febrile at 37.7°C and hemodynamically and respiratorily stable. Abdominal examination revealed a slightly distended abdomen with a painful right inguinal-scrotal swelling of inflammatory appearance. The transillumination test was negative on the right side, with the left testicle in place (Figure 2).

Biological tests showed normal renal function with hyponatremia at 129 mmol/l. Radiological imaging, represented by a standing thoraco-abdominal X-ray, revealed hydro-aeric levels in the groin and colon.

The diagnosis of a right inguino-scrotal hernia complicated by occlusion was made. The neonate was admitted to the operating room on a heated table after good fluid and electrolyte resuscitation. An incision was made in the right lower abdominal fold, followed by identification and careful dissection of the hernia sac. On opening, a necrotic distal portion of the appendix was revealed, with incarceration of a segment of the ascending colon and the presence of adjacent false membranes. Further exploration of the intestinal loops showed them to be viable, but a defect in the docking of the right colon was noted. Surgery consisted of appendectomy with reintegration of the herniated colon, followed by resection and closure of the hernia sac. The post-operative course was marked by resumption of intestinal transit after 24 hours, with abdominal collapse and disappearance of vomiting, the newborn had resumed his transit at the first day post operatiore, good clinical evolution, was declared discharged at the five day post operatiore.

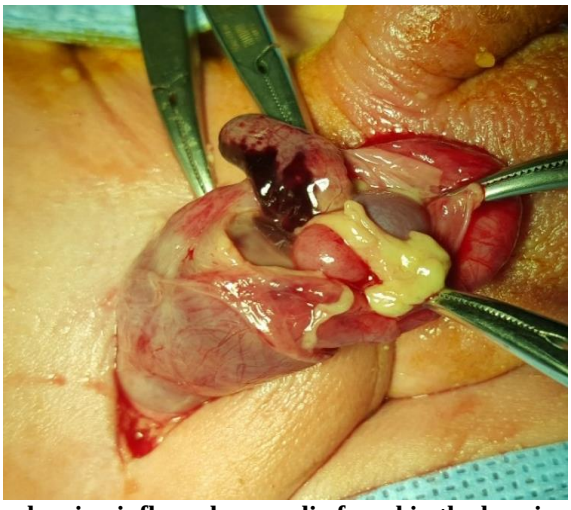

Figure 2: Preoperative picture showing inflamed appendix found in the hernia sac with some adhesions to sac

DISCUSSION

Amyand's hernia is an extremely rare occurrence in pediatric patients typically, the clinical presentation is that of a strangulated hernia with or without complications such as febrile occlusive syndrome [1, 6].

The pathogenesis of the presence of a vermiform appendix in a hernia sac is not clearly defined [7], which occurs in 0.1% of inguinal hernia cases [8–10]. The presence of the appendix in the hernia sac predisposes to the development of adhesions between its serous membrane and the hernia sac, resulting in an irreducible hernia [10]. Contraction of the anterolateral abdominal muscles leads to an increase in intra-abdominal pressure, causing compression and functional obstruction of the prolapsed appendix [11]. All these phenomena lead to irreducibility of the hernia, causing swelling with alteration of the microcirculation of the appendix wall, which can lead to perforation of the appendix and Amyand's hernia is difficult to diagnose clinically and is rarely diagnosed preoperatively. In a review of 60 cases over a 12-year period, only one case was diagnosed preoperatively [3]. The difficulty of diagnosis is due to the wide variety of symptoms presented by patients depending on whether the appendix is normal, incarcerated or perforated, the most common symptom being painful inguinal or inguinoscrotal swelling, whereas history and examination usually suggest an incarcerated hernia [14]. Fever and leukocytosis are inconsistent findings. Preoperative computed tomography (CT) has occasionally revealed the previously unsuspected diagnosis and is useful for early diagnosis, but is not routinely wused in clinical situations where a complicated hernia is suspected. Laermans et al have shown that the combination of CT and multiplanar reconstruction is the most useful technique to better visualise the appendix and its relationship with surrounding structures, thus helping to confidently make the correct diagnosis preoperatively [15]. Treatment of Amyand's hernia depends on the inflammatory state of the appendix. The practical approach to Claudius Amyand hernia depends on the appearance of the vermiform appendix in the hernia sac, and the clinical picture [16]. Appendectomy by inguinal herniotomy followed by closure of the peritoneovaginal canal is the ideal treatment for uncomplicated Claudius Amyand hernia. A median subumbilical laparotomy associated with the herniotomy is necessary in complicated cases. This attitude is codified by Lossanoff and Basson's classification into four types [17, 18]. This classification has been modified by the addition of a fifth type known as the Rikki modification [19, 20]. It should be noted that this modified Rikki classification does not take into account left Amyand's hernia, where a preventive appendectomy is recommended because, in the event of future appendicitis, there is a high risk of diagnostic error or delay [21, 22]. In an uncomplicated case, appendectomy followed by simple repair of the hernia through the same incision is recommended [3] In the presence of contamination, the use of synthetic mesh is absolutely contraindicated [11]. Laparoscopic reduction has also been described in the literature [23]. Appendectomy is a controversial procedure for most authors, as an organ containing faeces increases the risk of septic complications [3].

We examined the various approaches to the management of Amyand's hernia based on studies by four authors involving eight patients (Table 1).

Table 1: A literature review of reported cases of Amyand's hernia in children

Authors	Age	Sex	Preoperative diagnosis	Reached side	Skin Incision	Positive diagnosis	Treatment
CS *et al.*, 2022 [24]	18 months	M	Strangulated scrotal-inguinal hernia	Right	skin incision in the lower abdominal fold opposite the superficial opening of the inguinal canal	Per-operative	Appendicectomy and hernia sac closure
	10 months	M	Engorged scrotal-inguinal hernia				
	8 years old	M	Simple scrotal inguinal hernia				
	22 months	M	Simple scrotal inguinal hernia				
Dange *et al.*, 2013 [19]	3 years old	M	Simple scrotal inguinal hernia				
Fouda *et al.*, 2023 [25]	5 years old	M	Simple scrotal inguinal hernia				
Silvere *et al.*, 2022 [26]	12 months	M	Strangulated scrotal-inguinal hernia				
	3 years old	M					

CONCLUSION

Amyand's hernia is a rare condition that presents as a strangulated hernia. Diagnosis is mostly clinical, often unrecognized, and mostly intraoperative. Treatment consists of reduction of the contents of the hernia sac, possibly by surgical appendectomy, followed by ligation of the hernia sac.

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