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Case Report

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Amyand hernia with perforated appendix simulating as testicular torsion in a neonate: A case report

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KEYWORDS

Amyand hernia,
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ABSTRACT

Background: Amyand hernia is defined as the presence of the vermiform appendix within the hernial sac. A perforated appendix is an uncommon complication of Amyand hernia. Rarely it may simulate a testicular torsion.

Case Presentation: A twenty-five-day-old male infant presented with right-sided scrotal swelling, exhibiting symptoms of crying, irritability, and poor feeding over a two-day duration. The swelling was non-reducible, confined to the scrotum, and the spermatic cord showed thickening with erythematous scrotal skin. Following preparation, an inguinal incision was made, and an appendectomy and herniotomy were performed.

Conclusion: Amyand hernia, a rare type of inguinal hernia, poses challenges in preoperative diagnosis. The occurrence of a perforated appendix is especially rare in neonates with Amyand hernia. Surgical management entails appendectomy through an inguinal incision coupled with meticulous hernia repair.

INTRODUCTION

Amyand hernia is defined as the presence of the vermiform appendix within the hernial sac of an indirect inguinal hernia. [1-3] Claudius Amyand first described it in 1736. The incidence of Amyand hernia is about 1%, while the incidence of complicated hernia by acute appendicitis is only 0.1%, possibly occurring due to appendiceal incarceration within the hernial sac. [4] Males are more commonly affected than females, with a male-to-female ratio of 9:1. [5,6]

Clinically, it is difficult to differentiate Amyand hernia from incarcerated inguinal hernia, as it is not suspected preoperatively. Once the appendix becomes inflamed or perforated within the hernial sac, it poses a 14-30% mortality rate, compared to a 0.5-5% mortality rate for an inflamed or perforated appendix in its normal anatomical site. [7]

Herein we report a case of neonate presented with acute scrotum secondary to the Amyand hernia with perforated appendicitis that is simulated as testicular torsion.

CASE REPORT

A twenty-five-day-old full-term male neonate, delivered via normal vaginal delivery (NVD), presented with right-sided scrotal swelling, accompanied by crying, irritability, and poor feeding for two days before seeking consultation from the pediatric surgery department. The baby did not experience vomiting and had normal bowel motion.

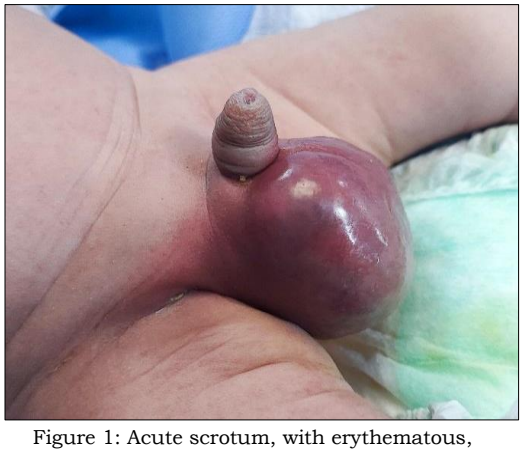

Figure 1: Acute scrotum, with erythematous, fluctuant right hemiscrotum

Upon examination, the baby appeared generally irritable and afebrile, with a nondistended abdomen. Locally, the right hemi-scrotum was observed to be tensely swollen, fluctuant, and tender, with a shiny red erythematous skin (Fig. 1). The swelling was non-reducible, confined to the scrotum, and the spermatic cord was slightly thickened.

An erect abdominal X-ray was performed, revealing a single air-fluid level in the right scrotal region, with normal gas distribution in the abdomen with no other air-fluid level (Fig. 2). The sonographic report suggested testicular torsion. Subsequently, the baby was kept nil per mouth, received IV fluid resuscitation, and was administered antibiotics. Preoperative laboratory investigations, including CBC (showing leukocytosis at 18×10^9/L), renal function tests, and virology screening, were conducted.

Figure 2: Erect abdominal X-ray showing a single air-fluid level in the right scrotum

Under general anesthesia and in the supine position, a transverse scrotal incision was made, revealing a release of air followed by watery stool. After complete stool evacuation, the testis was found in the upper scrotum without any evidence of bowel loop in the scrotum. An inguino-scrotal incision was then performed, and the spermatic cord, which was slightly thickened, was delivered. Dissection of the cord with separation of the hernial sac was undertaken, revealing a long appendix within the hernial sac, extending from the inguinal ring down to the fundus of the sac, with a perforated tip. A fibrous band was also identified between the tip of the appendix and the testis (Fig. 3). Appendectomy with herniotomy was performed, along with saline irrigation of the wound. A corrugated drain was left in, and closure was achieved using interrupted silk stitches.

Postoperatively, the patient continued to receive IV fluids, meropenem, metronidazole, and paracetamol infusion. Enteral feeding commenced the next day. The drain was removed on day 3 postoperatively, and the patient was discharged home a day later.

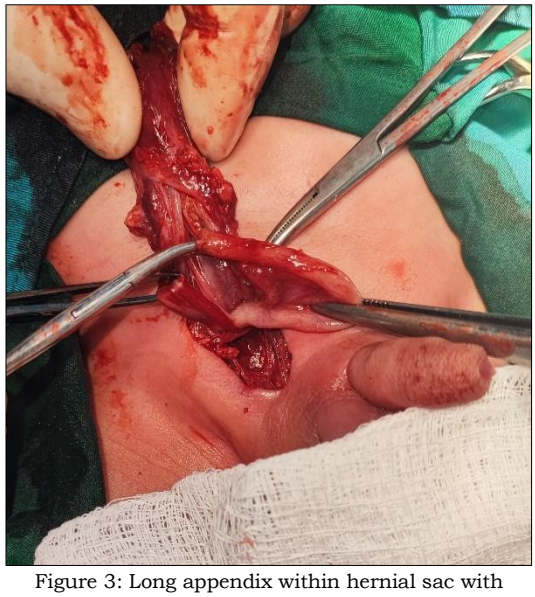

Figure 3: Long appendix within hernial sac with perforation at the tip pointed by mosquito

On day 5, a scrotal wound infection developed, prompting readmission to the hospital for an additional two days of parenteral antibiotics. The patient was discharged in stable condition thereafter. Inguinal stitches were removed on day 7 postoperatively.

DISCUSSION

Amyand hernia is typically diagnosed intraoperatively and requires a high index of suspicion for the diagnosis, particularly when faced with an irreducible inguinal hernia lacking the typical signs and symptoms of intestinal obstruction. [8] In our case, the preoperative diagnosis leaned towards a scrotal abscess (secondary to testicular torsion) rather than an obstructed inguinal hernia. This was supported by features such as erythematous and shiny scrotal skin, fluctuation, and a slightly thickened spermatic cord.

In instances where the appendix within an Amyand hernia becomes inflamed, the clinical presentation may imitate epididymo-orchitis, inguinal lymphadenitis, or testicular torsion. Such cases often face delayed diagnosis and treatment. [9] The pathophysiology of appendicitis and perforation in Amyand hernia remains unclear, with some authors proposing a decrease in blood supply during obstruction and incarceration or suggesting that the maneuver of manually reducing the hernia may lead to appendix inflammation and/or perforation. [10]

Perforated appendix in the context of Amyand hernia is more commonly observed in later childhood, with very few cases reported in the neonatal period. [11, 12] The presence of a narrow neck of the hernial sac, coupled with an impacted appendix, may prevent

stool leakage from the perforated appendix into the peritoneal cavity, thus obscuring typical clinical features of perforated appendicitis. In our case, there were no signs of peritoneal irritation or contamination, and the patient maintained a normal temperature.

Like the two reported cases, our case exhibited a fibrous connection between the tip of the appendix and the testis. This fibrous connection is believed to guide the passage of the vermiform appendix into the hernial sac with the assistance of the patent processus vaginalis. [13,14] A review of similar reported cases in the neonatal period, along with their clinical and operative details, has been summarized in Table 1. [15-20]

Table 1. A literature review of similar reported cases

Study name	Age & Maturity	Presenting features	Surgical approach	Operative findings	Surgical procedure	prognosis
Antonios Panagidis, et. al (2015) [15]	25 days Premature	Fecal discharge from scrotum (entero-cutaneous fistula)	Inguino-scrotal incision	Perforated appendix, which was adherent to the testis	Appendectomy and herniotomy	Well, no recurrence of hernia was reported
Ahmed Mohamed, et. al (2019) [16]	19 days Full-term	Persistent irritability, inguinoscrotal erythema, distended abdomen	Right upper quadrant incision	Inflamed, suppurative appendix incarcerated in the right internal ring	Appendectomy and purse string of internal ring	Well and stable through a three-month follow-up
Edoardo Guida, et.al (2020)[17]	15 days Full-term	Scrotal swelling and erythema, irritability, and poor feeding	Transverse scrotal and inguino-scrotal incision	Inflamed appendix inside hernial sac.	Appendectomy and herniotomy	Not documented
Vadyala Akshita Reddy, et.al (2021)[18]	3 days Extremely premature	Right scrotal swelling and erythema	Inguinal and scrotal incision	Perforated appendix with adherent tip to scrotal wall, with large amount of pus	Appendectomy and bilateral herniotomy	Well through one-year follow-up, with no evidence of hernia recurrence
Su Yeon Lee, et.al (2022) [19]	30 days Extremely premature	Right groin swelling, mild scrotal ecchymosis	Combined laparoscopy and right groin incision	Bilateral inguinal hernia, with incarcerated- -perforated appendix inside right inguinal ring with pus collection	laparoscopic left herniotomy, open right herniotomy with appendectomy	Well through a six-month follow-up, with no evidence of hernia recurrence
Khaled S. Abdullateef, et.al (2023) [20]	26 days Full-term	Tense, irreducible right inguinal swelling with skin erythema	Right inguinal exploration	Perforated appendix with dense attachment to hernial sac	Herniotomy and appendectomy	Well through a short period of follow-up

In conclusion, Amyand hernia stands as a rare manifestation of inguinal hernia, posing challenges in preoperative diagnosis. The occurrence of a perforated appendix within the context of Amyand hernia, particularly in neonates, is an infrequent complication. It may simulate other causes of acute scrotum. Surgical management typically entails performing an appendectomy through an inguinal incision, coupled with careful attention to hernia repair.

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