



Case Report

Perforated Appendix in Amyand Inguinal Hernia in a Neonate Presenting as Obstructed Oblique Inguinal Hernia: A Case Report and Review of Literature

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Abstract:

An Amyand hernia is defined as when the appendix is trapped within the sac of an inguinal hernia through a patent vaginal process. It was first described by Claudius Amyand, in 1735 during an appendectomy of a perforated appendix inside a hernial sac of an 11-year old child. Amyand hernia rare, constituting less than 1% of inguinal hernias. Amyand hernia is 3 times more likely to be diagnosed in children compared to adults due to the patency of vaginal process. Appendicitis or its perforation in Amyand hernia, complicate 0.1% of cases. Other variants include the presence of ileum, caecum or both in addition to the appendix. Isolated acute appendicitis in neonates is extremely rare, with a high mortality rate, and higher among those within Amyand hernia up to 30%. Perforated appendix in Amyand hernia is difficult to diagnose, as its clinical picture is not specific. We report a neonate 26 days old with perforated appendix in an Amyand inguinal hernia who presented by a picture of obstructed oblique inguinal hernia. He underwent emergency open inguinal exploration, herniotomy and appendectomy. Despite the high mortality rate of Amyand hernia due to be peritoneal spread of sepsis, the boy survived complication free. Perforated appendix in Amyand hernia is exceptionally rare, and can present in the early neonatal period, its diagnosis and outcome rely on high index of suspicion and prompt surgical intervention.

Level of Evidence of Study: IV (1).

Keywords: inguinal hernia; Amyand hernia; perforated appendix; neonate.

Introduction

Amyand hernia is defined as the entrapment of the appendix within the sac of an inguinal hernia (2–4). It is rare, and is mostly clinically silent (3). Yet if the appendix in Amyand hernia is inflamed and or perforated; it is an emergency. Clinically it is indistinguishable from incarcerated inguinal hernia, as Amyand herniation of the appendix is not suspected pre-operatively. As once it is inflamed or perforated in the tight space of the inguinal hernia it poses 14-30% mortality, compared to the 0.5-5% of mortality in the inflamed or perforated appendix in its anatomical place (5). Despite being very rare, it is mostly a disease of older children rather than adults. We aim to describe our experience in a 26-day-old neonate with perforated appendix in Amyand hernia.

Case Presentation

A full-term 26-day-old male infant was brought by his mother to the outpatient clinic presenting with an accidentally discovered 2-day history of right inguinal swelling and 1-day history of being tense and irreducible. The mother gave history of non-bilious vomiting and normal defecation. On physical examination there were tenderness over the irreducible swelling and the skin overlying was edematous and erythematous. The neonate was vitally stable with no fever or features of sepsis. Preoperative complete blood picture showed hemoglobin of 13 mg/dl and total leukocytic count of 21,000/dL. Preoperative imaging was deemed unnecessary, and the patient was diagnosed with an obstructed oblique inguinal hernia. During inguinal exploration for hernia repair, the sac contained viable caecum and perforated appendix which was attached to the sac but there were no bands. (Figure 1). The neonate underwent a herniotomy and an appendectomy. On day 4 post operatively, day 30 of life, the patient was vitally stable on room air and was taking full bottle feeds. The abdomen was lax, and the scrotum was elevated. Antibiotics were prescribed and the patient was discharged by day 5. Follow up was uneventful with a right inguinoscrotal edema that resolved spontaneously.



Figure 1. Amyand hernia as evident during inguinal exploration for obstructed hernia repair. The sac contained viable caecum and perforated appendix attached to the sac.

Discussion

Obstructed inguinal hernia is an emergency based on a clinical diagnosis. We did not anticipate perforated appendix in Amyand hernia in our neonate, as it is an operative finding. It is interesting though that the neonate was not feverish and did not present in sepsis, despite the raised total leukocytic count. The smooth march, uneventful recovery and outcome seems to related to the prompt surgical interference. Amyand hernia does not need a different surgical approach, hence there is no recommendation to undergo imaging (6) prior to intervention once the clinical diagnosis of obstructed hernia is made.

A non-complicated case of Amyand hernia may be asymptomatic or may present with nonspecific symptoms that mimic orchitis, testicular torsion, or enterocolitis (7). However, in cases of appendicitis the diagnosis of Amyand hernia is challenging and requires a high index of suspicion. Clinically, the preoperative diagnosis of Amyand hernia is difficult, and most cases present as incarcerated right inguinal or inguinoscrotal swellings so awareness of the surgeon about this rare condition as well as the clinical finding of irreducible hernia without the cardinal symptoms of intestinal obstruction raises the possibility of Amyand Hernia (8).

The general manifestations like fever, vomiting, and abdominal distention depend on the condition of the vermiform appendix whether inflamed or perforated, and the clinical picture may be obscure due to the narrow neck of the sac of the herniation which limits the spread of inflammation and peritoneal irrigation (9), as evident in our patient in which the inflammation did not spread to the peritoneal cavity and was confined to the scrotum, mimicking a strangulated hernia, otherwise it would have spread within the abdominal cavity as described in infants with normal location of appendix. Again the clinical picture is heterogenous, as there is a recent report of appendicitis in a 19-day-old neonate who presented by inguinoscrotal edema and erythema without palpable inguinoscrotal mass. The differential diagnosis of inflamed



appendix in Amyand hernia includes not only necrotizing enterocolitis, torsion of the testis and epididymo-orchitis (10), but also intestinal obstruction which are all emergencies. The non-specific challenging clinical picture results in delays in diagnosis and surgical exploration that increases morbidity and mortality up to 30%. Diagnosis of appendicitis in neonates is reported to be extremely rare with less than 200 reported cases (11), it is not clear if it is underdiagnosed, as its clinical picture might mimic conditions that are medically managed such as sepsis, necrotizing enterocolitis and ileus. Moreover, in neonates it can be confused by intussusception where air enema decompression and delayed repeat enemas might delay the operative intervention up to 12 hours (12), and compromise the outcome.

Being a male carries higher incidence of Amyand hernia. Male to female incidence is 9:1 (13, 14). During the operation we did not encounter fibrous bands or connections, contrary to some reports that suggested a presence of fibrous connection between the appendix and the testis that act as a guidance to the passage of the appendix with the aid of patent processus vaginalis into inguinal hernia (15, 16). The age at which perforated appendix presents in Amyand hernia is variable, commonly in later childhood, yet very limited case reports describe cases as early as 3 weeks, in preterms and full terms and as late as 68 years (17, 18). Other reports proposed that Amyand hernia results from repeated microtrauma, adhesions, and recurrent acute inflammation which occurs due to migration of the appendix within the hernia sac (19). We did not come across bands or fibrosis in our case. It seems that sliding of the appendix in the inguinal hernia sac might be related to the short mesoappendix that allows the appendix to freely float in peritoneal cavity (20).

In cases of Amyand hernia with appendicitis, it is accepted that appendectomy should be performed but the controversies about Amyand hernia with normal looking appendix are still present. Many authors believe that the normally looking appendix which is incidentally discovered during surgery without any signs of inflammation should not be removed, and prophylactic appendectomy is not necessary (8). Among adults Amyand hernia surgical management relies upon its class according to Losanoff and Basson (21), which is not the case for children.

Our case was managed by open surgery. Advances of minimal invasive surgery in the recent years made laparoscopic congenital inguinal hernia repair a feasible and simple procedure. It has the advantages of clear visualization of the internal ring in both sides and exploration of the abdomen and pelvis, especially the pelvis which is better visualized using minimal invasive approach than open approach, reduction of the irreducible contents under direct vision and detection of the vascularity of the reduced viscera, precise suturing of the peritoneum at the internal ring, and meticulous manipulation of the vas deference and testicular vessels to avoid injury of both structures (22, 23). It remains to be seen if laparoscopic procedures offer advantages in Amyand hernia surgical management. Advances in artificial intelligence in deep learning in surgery is rapidly evolving and might prove to be valuable in pre-operative diagnosis of Amyand hernia (24).

Conclusion

Amyand hernia is a rare type of inguinal hernia, present more in children compared to adults due to patency of vaginal process. Once the appendix within Amyand hernia is inflamed, there is high risk of morbidity and mortality. It masquerades as obstructed hernia. Prompt surgical management is lifesaving. It can occur at early neonatal period, without constitutional manifestations. There is a need for consensus and guidelines for diagnosis of perforated appendix in Amyand hernia.

Author Contributions:

All authors shared in the work, read and agreed to the published version of the manuscript.

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CONFLICT OF INTEREST

The authors declare no conflict of interest in connection with the reported study. Authors declare veracity of information.



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