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Amyand's hernia with perforated appendix and scrotal abscess in a premature newborn

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Abstract (69 words)
An Amyand's hernia is an unusual diagnosis in children and even rarer in neonates. Perforation of the appendix inside the hernia sac is also extremely unusual. The diagnosis of an Amyand's hernia is difficult as presenting symptoms can also be attributed to an isolated inguinal hernia, noncommunicating hydrocele, or testicular torsion. We present a case of an Amyand's hernia with perforated appendix and scrotal abscess in a premature newborn.

Key Words
Amyand hernia
Perforated appendicitis
Pylocele
Scrotal abscess

Introduction
Amyand's hernia is an inguinal hernia containing the appendix. Amyand's hernia is a rare condition, occurring in less than 1% of pediatric inguinal hernias.[1] While cases of Amyand's hernia are unusual in children with an incidence of 0.42%, a perforated appendix in the inguinal hernia sac is even more uncommon, occurring in less than 0.1% of all Amyand's hernias.[1] Additionally, with less than 0.012% of appendicitis cases occurring in the neonatal period, the concurrent presentation of appendicitis with Amyand's hernia is exceedingly rare.[1] Signs and symptoms of intestinal obstruction are present in the majority of Amyand's hernia cases.[1] We

report an unusual case of an Amyand's hernia containing a perforated appendix in a preterm neonate who presented without clinical signs of obstruction.

Case Report

A 2200g male infant was born at 28 weeks gestational age and admitted to the neonatal intensive care unit (NICU). At 5 weeks of age, he developed right groin swelling. He continued to stool regularly and tolerating feeds. He did not have leukocytosis. Of note, he was receiving empiric antibiotics for presumed respiratory infection. His physical exam revealed a reducible firm lump in the inguinal canal and fluid around the right testicle without overlying skin changes at the groin. Mild ecchymosis was present at the inferior lateral aspect of the right testicle that remained stable. A scrotal ultrasound demonstrated the right testicle within the right hemi-scrotum with slight hypervascularity, enlargement, and additional avascular echogenic material mainly in right scrotal sac thought to represent hemorrhage or a complex hydrocele. Given these findings, the initial diagnosis was a reducible inguinal hernia with an associated hydrocele. While the infant never demonstrated any clinical signs of obstruction and the hernia remained reducible, he underwent repeat scrotal ultrasound three and eight days later because of persistent edema and scrotal discoloration. These studies re-demonstrated flow to right testicle, extra-testicular complex fluid collection with increased component of simple fluid, bilateral scrotal thickening with hyperemia of scrotal sac lining, and ultimately, bowel-containing right inguinal hernia and right inguinal lymphadenopathy (Figure 1). At this time we became concerned for an incarcerated hernia. We performed a diagnostic laparoscopy with planned repair of the right inguinal hernia. At the time of surgery, he was 7 weeks old (corrected gestational age of 35 weeks) and weighed 2535g.

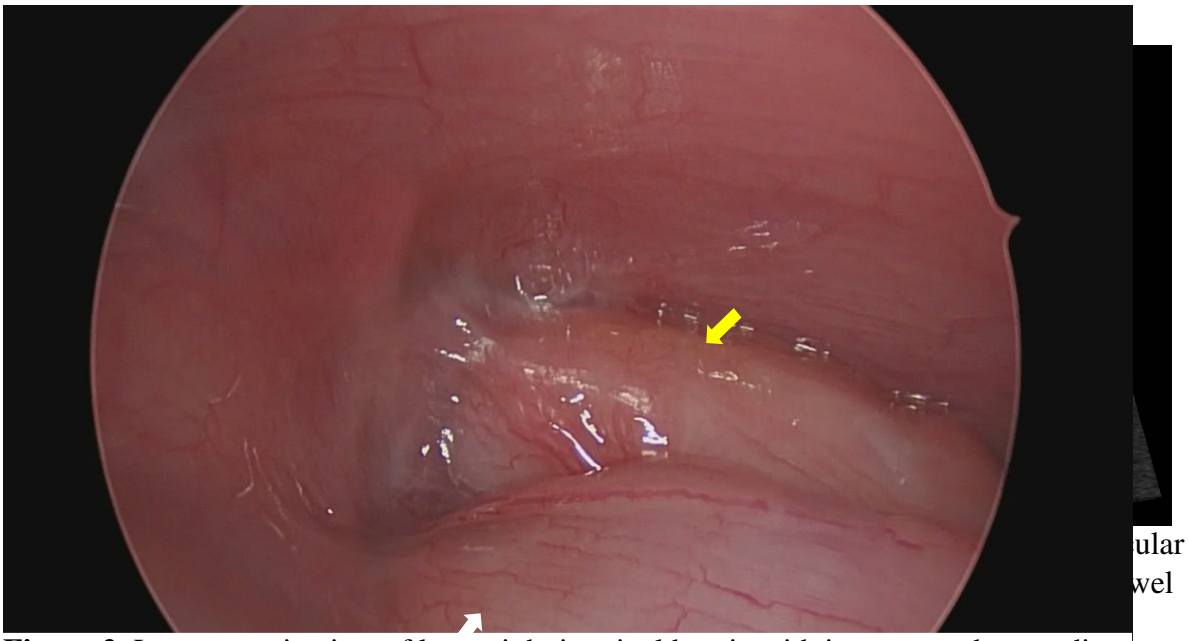

Figure 2. Laparoscopic view of large right inguinal hernia with incarcerated appendix (yellow arrow) and tethered cecum (white arrow) in the right lower quadrant.

Diagnostic laparoscopy identified bilateral indirect inguinal hernias. The left side contained no bowel contents and was repaired primarily with high ligation of the ring using the spinal-needle technique without complication.[2] On the right side, however, it appeared that the appendix was incarcerated into and closing off a large inguinal defect (Figure 2). The cecum was tethered to the lateral wall adjacent to the inguinal ring with a moderate amount of surrounding inflammation in the right lower quadrant. A right groin incision was made to better understand the anatomy and more safely identify the testicular structures. A large scrotal abscess was encountered and drained. The appendix was carefully separated from the testicle and cord. Care

was taken to ensure that the testicle remained perfused and viable. The appendix demonstrated evidence of chronic appendiceal incarceration, perforation, and abscess (Figure 3). An open appendectomy was performed followed by a high ligation of the right hernia sac with primary closure of the conjoint tendon to the inguinal ligament (Bassini repair). Given the chronically infected field, a vessel loop drain was placed through the inguinal incision and out a counter incision in the scrotum.

The patient recovered well after surgery without complication. Intra-operative scrotal cultures grew *Enterobacter cloacae*, *Klebsiella oxytoca* and *Propionibacterium*. He received piperacillin/tazobactam until vessel loop removal on post-operative day 5. He was discharged home one month later once his prematurity-related respiratory status improved. There were no hernia recurrences or surgical site infection on follow-up at 6 months.

Discussion

We report an unusual case of an Amyand's hernia with appendiceal perforation and scrotal abscess which was initially believed to be a reducible inguinal hernia with an associated complex hydrocele. An Amyand's hernia with a perforated appendix is extremely rare, estimated to be less than 0.1% of all Amyand's hernia cases.[1] A study of 7,138 pediatric patients with inguinal hernias reported only 30 cases of an Amyand's hernia, 5 of which were inflamed and 2 perforated.[1] Another study of 4,498 children with inguinal hernia reported 46 cases of an Amyand's hernia, of which 9 appendices were inflamed and 1 perforated.[3] Another series of 1,090 children with inguinal hernias reported 12 patients with Amyand's hernia, 10 of whom had an inflamed appendix.[4] Two case reports documented Amyand's hernia with perforated appendix

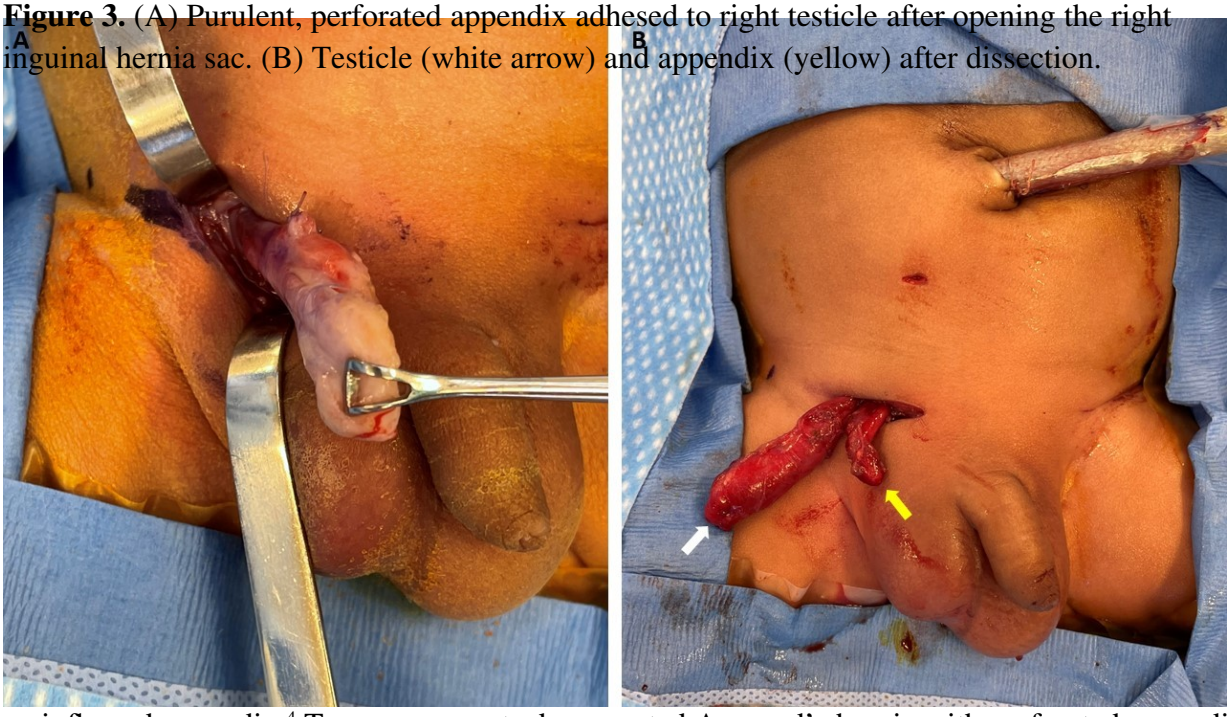

Figure 3. (A) Purulent, perforated appendix adhesed to right testicle after opening the right inguinal hernia sac. (B) Testicle (white arrow) and appendix (yellow) after dissection.

in neonates.[5,6] Our case is the first to our knowledge to report perforated appendicitis with associated pylocele in an Amyand's hernia in a preterm neonate.

The diagnosis of an Amyand's hernia can be challenging to make. The differential diagnosis includes an incarcerated inguinal hernia, a noncommunicating hydrocele, testicular torsion, epididymitis or orchitis. While erythema, tenderness, irreducibility, and intestinal obstruction are signs that warrant urgent surgical exploration, a seemingly reducible hernia without obstruction made it difficult to make the diagnosis in our case. In our patient, the appendix had likely perforated early on, which then falsely made the hernia seem reducible on physical exam. He probably was not obstructed due to the sequestration of the appendix and associated pylocele within the scrotum. Additionally, the antibiotics the patient was concurrently receiving further masked the signs of soft tissue inflammation and appendicitis.

Most cases of Amyand's hernia are discovered intraoperatively. Out of the 12 cases described by Cankorkmaz et al, only one patient was diagnosed preoperatively by ultrasound.[4] Other imaging modalities including computed tomography (CT) scan have been used more frequently in adults for diagnosis. However, a retrospective review of 6 adult cases concluded that CT scans were nondiagnostic, and recommended laparoscopy for definitive diagnosis.[7] If cross-sectional imaging is concerned for neonates, CT scan should be used judiciously given the concern for developing radiation-related cancer.[8,9]

Given the rarity and difficulty of diagnosing this disease, correct and timely identification of an Amyand's hernia depends heavily on physicians' awareness of this possibility.[7] Given the 15-30% increase in the mortality of Amyand's hernia with perforated appendix compared to that of normal Amyand's hernia, suspicion must remain high.[7] An Amyand's hernia with perforated appendix should be on the differential in neonatal patients with complex hydrocele, even when hernia appears reducible and without obstruction.

Conclusion

An Amyand's hernia is a rare condition in which the inguinal hernia contains the appendix. This condition can occur even in neonates, and can be further complicated by perforated appendicitis and scrotal abscess. If incarceration and perforation occur, proper diagnosis is challenging as symptoms can also be consistent with a reducible inguinal hernia with hydrocele. It is important for neonatologists and pediatric surgeons to be aware of this diagnosis even in cases that do not present with the typical symptoms of obstruction.

Patient consent

Written informed consent to publish this case report was obtained from the patient's parent.

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All authors attest that they meet the current ICMJE criteria for Authorship.

Declaration of competing interest
The authors declare that they have no known competing financial interest or personal relationships that could have appeared to influence the work reported in this paper.

Colour should be used for any figures in print.

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