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Amyand's Hernia in an Infant: A Case Report and Review of Literature

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Abstract

Introduction: Inguinal hernia is a common surgical disease among a pediatric population with patent processus vaginalis being the major risk factor. Omentum and small intestine loops are the often-encountered hernia sac contents, however in a rare instance (1% of cases), a vermiform appendix is found in the sac, and it is referred as Amyand's hernia (AH). The appendix can be normal, inflamed, strangulated, or perforated.

Case presentation: 7-week-old male baby with 2-week history of right groin swelling, excessive crying and restlessness. He was clinically stable with irreducible right inguinoscrotal swelling. An ultrasonography confirmed a 1.5 cm abdominal wall defect with bowels in the sac. A clinical diagnosis of incarcerated inguinal hernia was made, and the patient was prepared for surgery. A standard herniotomy incision was done, hernia sac was found and opened and appendix and cecum were found. The contents were reduced, the sac was ligated and transected, and the incision was closed in layers. The post-operative follow ups were uneventful.

Discussion: AH is a rare condition, with estimated prevalence of 0.19% to 1.7% and 0.1% occurrence of appendicitis. The clinical presentations of AH include painful groin swelling, inguino-scrotal erythema, tenderness, and features of obstructed or strangulated inguinal hernia. Diagnosis is often made during surgery as it is exceedingly difficult to diagnose it pre-operatively as it resembles inguinal hernia, and it can mimic testicular torsion or inflammation in case of erythema and tenderness. The surgical approach depends on the status of appendix, and it can be reduction of appendix or appendectomy if inflamed.

Conclusion: Although rare, an index of suspicion should be kept in all inguinal hernia repairs. The appendix should be preserved if normal as with emergence of novel surgical technologies, the appendix can be of significant use especially for the children.

Keywords: Inguinal hernia; Amyand's hernia; Case report; Herniotomy; Inguino-scrotal hernia.

Abbreviations: AH: Amyand's hernia; CT: Computed Tomography; WBC: White Blood Cells.

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Introduction

Inguinal hernia results from a defect in the anterior abdominal wall with abdominal contents herniating into the groin [1]. The location of the defect in relation to the inferior epigastric vessels has resulted in a common classification of inguinal hernias into direct and indirect types. The direct (medial) inguinal hernia occurs medial to the inferior epigastric vessels while indirect (lateral) inguinal hernia is medial to these vessels and passes through the inguinal canal and can descend into the scrotum [2]. In children, indirect inguinal hernia is the most common type with patent processus vaginalis being the major risk factor. A major risk factor for occurrence of inguinal hernia in adults is increased intraabdominal pressure. Increased intraabdominal pressure results from strenuous exercises, heavy weightlifting, chronic coughing, bladder outlet obstruction, and chronic constipation. Other risk factors include cigarette smoking, old age, connective tissue disorders, pregnancy. Lifetime risk of developing inguinal hernia is higher in males as compared females (27% and 3% respectively) [2]. Small intestine is the most commonly encountered hernia content but in rare cases, about 1%, a vermiform appendix can be found in the hernia sac [3]. Diagnosing Amyand's hernia preoperatively is difficult even with the use of CT-scan and may prove to be an intriguing intraoperative finding. [4] Treatment of Amyand's hernia depends on the status of the appendix, it can be reduced if normal or resected if inflamed or perforated [1]. We aim to present a rare case report of Amyand's hernia in an infant and the work has been reported in line with the SCARE criteria [5].

Case presentation

Seven weeks old male baby who presented to our center with the chief complaint of rightsided inguino-scrotal swelling for two weeks. The swelling was initially reducible and prominent only when the baby coughs or yawns but two days prior to admission, the swelling became non-reducible. This was associated with excessive crying and restlessness; however, there was no reported history of fever, no change in bowel habits, no abdominal distension, no vomiting, and breastfeeds well. The child was delivered at 39 weeks of gestation, weighing 2.4 kg, and cried immediately after delivery.

On examination: Alert, active baby, afebrile, not pale, not jaundiced, not cyanosed. Temperature – 36.6^0C, Pulse rate – 108 beats/minute, respiratory rate – 28 breaths/minute SPO_2 – 99% in room air. Per Abdominal & groin examination: Normal abdominal contour, non-tender firm swelling in the inguinal region and right scrotum about 6 cm x 5 cm, non-reducible, palpable testicles, and negative illumination test. Laboratory tests: Full blood picture was normal (WBC – $7.4x10^9$/L, hemoglobin – 13.1 g/dL, Platelets – $342x10^9$/L), serum electrolytes were within normal ranges (Sodium – 137 mmol/L and potassium – 4.2 mmol/L).

Scrotal ultrasonography: There was about 1.5 cm defect with herniation of bowels into the right inguinal canal and scrotum, with positive peristalsis. The testes appear normal in size and echotexture, no focal lesions, with normal intratesticular flow. Epididymis and spermatic cord thickness appear normal on both sides.

A clinical diagnosis of incarcerated right inguino-scrotal hernia was made, and the patient was planned for emergency herniotomy.

Surgery details: Under general anesthesia, a transverse right groin incision was made along a skin crease, the inguinal canal was opened, hernia sac was identified and separated from the spermatic cord. Hernia sac was opened and an uninflamed normally appearing vermiform appendix and cecum were found in it as shown in Figure 1. The cecum and the appendix were carefully reduced into abdominal cavity, hernia sac was suture ligated and transected, and the incision was closed in layers. An intraoperative diagnosis of Amyand's hernia was made.

Follow up: The patient recovered well from surgery and was discharge home after 3 days. He was seen after two weeks in outpatient clinic where the wound had healed with no complications.

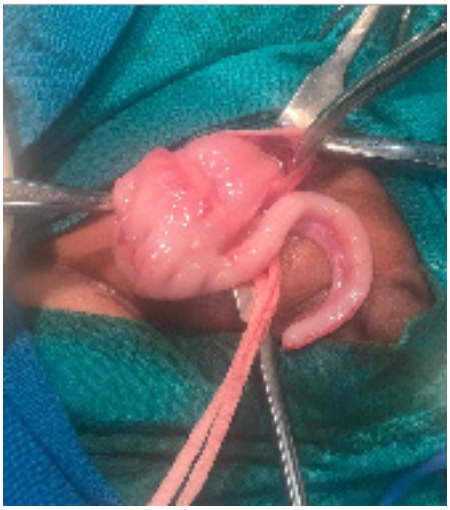

Figure 1: Intraoperative image showing the contents of the hernia sac which were cecum and a normally appearing appendix.

Discussion

As described earlier, Amyand's hernia is when an inflamed or normal vermiform appendix is found in the inguinal hernia. It was first described by Claudius Amyand in 1735 where he successfully operated on a boy with inguinal hernia which was inflamed and had a pin and a stone in it [2]. For a period of over 200 years, there has been a debate on what should be Amyand's hernia as it was first described as inguinal hernia containing an inflamed appendix, but since then it has been described even in absence of appendicitis, perforation, or ischemia [6]. AH is rare, with estimated prevalence of 0.19% to 1.7% and 0.1% occurrence of appendicitis. It is reported to be 3 times more in children as compared to adults [1]. Although exceedingly rare, a left sided Amyand's hernia has been reported and it is postulated to result from a free-floating cecum, gut malrotation, and situs inversus [3,7]. Occurrence of appendicitis in children less than 2 years is about 2% with incredibly low occurrence in neonates (about 0.012%). Although appendicitis is less prevalent in neonates, AH has a bimodal distribution as it is more reported in premature neonates, infants, and postmenopausal women [8,9].

In a series of 30 patients, Kaymakci et al reported a painful inguinal or inguino-scrotal swelling to be the most common presentation of Amyand's hernia [8]. Majority of case, like our case present with features of obstructed or strangulated inguinal hernia. Among the few cases of inflamed vermiform appendix in the

inguinal hernia, majority do not have systemic manifestations of appendicitis [2]. Other presentations like groin tenderness, erythema, and irreducibility of contents have been reported in the literature [6]. Our patient presented with a painful irreducible groin swelling. Diagnosis of AH preoperatively can be exceedingly difficult particularly due to its rarity, and similarity with inguinal hernia. When there is appendicitis, it can present like a testicular inflammation or torsion, hence difficult to diagnose [6]. The use of CT-scan can significantly increase the preoperative diagnosis of AH, but it is rarely used in clinical practice as ultrasonography is usually adequate for diagnosing inguinal hernias. Laparoscopic assisted hernia repair in children is in practice and can be particularly useful for both diagnosing and treating AH [10].

The mainstay method of treating AH is surgery, and the diagnosis is mostly made intraoperatively. The surgical approach will depend on the status of the appendix. Most authors suggest reducing the appendix if normal or perform appendectomy and repair hernia simultaneously if inflamed, ischemic, or perforated. Prophylactic appendectomy in normal appendix in AH, is discouraged because appendix can be potentially used later in biliary reconstruction, antegrade enemas, and urinary diversion [1,7,8]. In our case, the vermiform appendix appeared normal, so it was just reduced with no complications seen during the follow up period.

Conclusion

Amyand's hernia is a rare condition and easily misdiagnosed as strangulated inguinal hernia, especially in a pediatric population. The diagnosis if often made intraoperatively, hence a high index of suspicion should be made. For this reason, a hernia sac should be opened and visualized before reduction of the contents and ligation of the sac. Avoidance of unnecessary appendectomy is highly suggested as novel surgical technologies might expand the use of appendix in the future, especially in a pediatric population with longer life expectancy. More research is highly encouraged for standardization of care and classification of AH because currently the approach is primarily dictated by a surgeon's preference.

Declarations

Funding: There was no funding for this work.

Conflict of interest: No conflicts of interest.

Ethical statement: Ethical review board ethical clearance is not needed for case reports in our institute; however, permission was granted by the head of the neurosurgery unit.

Parental consent for minor: Written informed consent was obtained from the patient's parents/legal guardian for publication and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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