Introduction

We report a 3-year-old child who presented with features of strangulated left inguinal hernia containing perforated cecum and appendix. Laparotomy revealed associated abnormal anatomy. This is the second reported case of the presence of cecal perforation in left Amyand’s hernia in pediatric age group.

Case Report

A 3-year-old term born male was brought by his parents to the emergency department for inconsolable crying for the past 6 hours. According to the parents, the patient had a small reducible swelling in the left groin first noticed around 6 months of age which had become painful and non-reducible for the past 5 days. On examination, a tense, tender, warm, indurated, erythematous, minimally fluctuant, transilluminant left-sided inguinoscrotal swelling was seen [Figure 1]. The scrotal rugosities were lost, left-sided testis could not be palpated, and the penile tissue was buried under skin with only prepuceal covering visible [Figure 1]. Clinically, a strangulated inguinal hernia with reactive fluid in the hernial sac was suspected. X-ray abdomen erect view showed no air-fluid level or free air in the peritoneal cavity or hernial sac. Ultrasonography (USG) demonstrated the presence of bowel loops with free fluid with internal echoes in the left scrotal sac with well-vascularized bilateral testes. On emergency exploration, initially, a left inguinal crease incision starting at the superficial inguinal ring was given. The incision relieved the tissue pressure around the penis and only then the prepuce and penile skin could be retracted and penile meatus visualized which was catheterized. Hernial sac was thick and densely distended. When it was opened, a gush of air came out along with fecal matter containing fluid. Sac contained perforated cecum and a normal looking appendix as its contents [Figure 2]. A decision to explore the patient...
Hernias occur in 3%–5% of all infants, more so in premature infants. Hernias, if left unattended, may progress to incarceration, obstruction, strangulation, or perforation which may convert this easily treatable surgical condition into potentially life-threatening situation. The term “Amyand’s hernia”, first reported by Claudius Amyand, is used for describing the presence of non-inflamed appendix, inflamed appendix, or perforated appendix in an inguinal hernia. “Amyand’s hernia”, can appear on the left side rarely, and only 15 pediatric cases have been described in the literature. All of the patients were males whose ages ranged from 2 to 18 months. Our patient is thus the 16th case and oldest patient by age among the reported cases of left “Amyand’s hernia”, in pediatric population.

The presentation in a case of left Amyand’s hernia can be simply a reducible swelling in the groin, or it can present as an irreducible swelling with or without features of obstruction. History and clinical examination is not enough to confirm the presence of appendix in hernial sac. Pre-operative imaging studies (especially a contrast-enhanced computed tomography) can be useful for confirming the presence of appendix and in diagnosing the presence of situs inversus, intestinal malrotation, or a mobile cecum as possible underlying anomalies, especially in the left-sided cases. The indications for a contrast-enhanced computed tomography (CECT) in a stable patient in such a case may include demonstration of appendix (a blind-ending tubular structure) in hernial sac in USG or demonstration of cecum in hernial sac in a pre-operative contrast study as demonstrated by Yoneyama et al. in their case.

Management is essentially surgical and it is based on intraoperative findings. On exploration, if appendicitis with or without abdominal sepsis or perforated appendix is present, appendectomy is essential. Appendectomy may also be done if the appendix appears normal, especially in case of the left-sided Amyand’s hernia as the cecum is mobile or the patient may have situs inversus or intestinal malrotation. This prevents diagnostic dilemma if appendicitis occurs in such a patient in future as it would have an atypical clinical presentation. Yoneyama et al. in their review of 14 cases of left Amyand’s hernia noted that appendectomy was performed in 8 cases; however, they did not perform appendectomy in their own case. One case was missed by Yoneyama et al. in their review of literature that is the case reported by Gupta and Sharma et al. which also had an inflamed appendix, and they also performed appendectomy. Appendicitis was noted in 5 of the 15 pediatric patients with left-sided Amyand’s hernia. Some authors say that if the appendix appears normal on clinical examination, it should not be resected out as it increases risk of surgical site infection, possibility of hernia recurrence, and nonavailability of appendix as a source for describing the presence of non-inflamed appendix, inflamed appendix, or perforated appendix in inguinal hernia.
of urinary conduit (Mitrofanoff appendicovesicostomy), if the patient ever requires it.\(^1\)

Usually, inguinal approach is enough to manage most of these cases; however, sometimes, a switch to formal laparotomy may be required as in our case. We took a decision to switch to laparotomy as it was not possible to return the hernia contents back to the abdomen due to the abnormal anatomy we encountered and to rule out situs inversus, intestinal malrotation, and mobile cecum and ascending colon. Our patient is the second case of a perforated cecum in left inguinal hernia in a child. The first case was reported by Singh et al.,\(^5\) but they did not perform appendectomy and primary repair of cecal perforation was done. We advocate midline bowel exploration in all patients of left Amyand’s hernia so that underlying anomalies can be ruled out except if associated anomalies have been previously ruled out by pre-operative contrast enterography or CECT.

Most cases of left Amyand’s hernia were due to mobile cecum, one case was due to situs inversus,\(^3,4\) but none of the reported case was due to intestinal malrotation. Our patient definitely had mobile cecum and ascending colon but also some degree of bowel malrotation due to the omental band as the bowel was rotated at the wide mesenteric base and present in the left iliac fossa. It also caused rotation of bowel on mesenteric axis; however, due to wide mesenteric base, there was no clinically apparent vascular compromise. We could not find any similar abnormal anatomy described in any of the embryology or anatomic variations textbook or during online literature search.

Although free air in hernia sac has been reported in some case reports,\(^6\) a specific term has never been used for this so we propose usage of the term “pneumohernios” (Gk., pneumon - lung; air; Gk., hernios - split, rupture) for clinical, radiological, or per-operatively demonstrable free air in any hernial sac as was evident in our case.

To conclude, in rare cases, hernias may present with great surprises. This is not only due to their content but also for the complexity in management. Thus, if an emergency exploration of the left inguinal hernia reveals an appendix and cecum; then, one must have a high index of clinical suspicion for a rotation–fixation anomaly of bowel or situs inversus and a laparotomy may be indicated to concomitantly manage the same.

**Declaration of patient consent**

The authors certify that appropriate patient consent was obtained.

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**Conflicts of interest**

There are no conflicts of interest.

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