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Management of Amyand's hernia in children: should appendectomy be mandatory or not?

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Abstract

Background: Amyand's hernia (AH) is defined as protrusion of the vermiform appendix within the sac of the inguinal hernia and usually misdiagnosed as obstructed hernia. It is more common in pediatric patients but there are controversies about dealing with the normally looking appendix in AH. The aim of this study is to present our experience in the management of AH and highlights the difference in dealing with normally looking appendix between pediatric and adult patients. Patients' demographics, intraoperative, and postoperative outcome were reported and analyzed.

Results: This is a retrospective analysis of 12 pediatric patients aged between 15 days and 5 years presented with AH and operated in tertiary referral hospital over 10 years. This study included 10 males and 2 females. The median age at the operation was 7 months. Eleven patients were operated using conventional technique, and only one patient was operated using laparoscopy. In 11 patients, the appendix was looking normal and was reduced without appendectomy, and the hernia was repaired. Appendectomy was done in one patient presented with AH containing inflamed appendix. The postoperative course was uneventful in all cases.

Conclusions: There is a difference in the management of pediatric and adult patients presenting with AH, and appendectomy is not mandatory in normally looking appendix in pediatric patients.

Keywords: Amyand's hernia, Appendectomy, Children, Management

Background

Amyand's hernia (AH) is a unique condition due to protrusion of a vermiform appendix in the sac of the inguinal hernia [1, 2]. It was discovered and firstly described by Claudius Amyand in 1735 when he managed a boy aged 11 years old presented with an inguinal hernia containing inflamed appendix with fistula between thigh and scrotum [3, 4]. The prevalence of Amyand's hernia is less than 1% (0.4% and 0.6%)

according to the literature while the prevalence of acute appendicitis in amyand's hernia reached 0.1% [5, 6]. It is three times more common in pediatric patients [7]. It is more common in males, and it has a bimodal age distribution in neonates and geriatric patients above 70 years old [8, 9].

Amyand's hernia presents as inguinal or inguino-scrotal swelling, and the preoperative diagnosis is uncommon as it is usually mistaken as incarcerated or strangulated hernia and so, it usually represents a surprise during surgery [10, 11]. Sonographic diagnosis of appendix within the hernia sac was reported in the

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literature [12, 13]. Abominopelvic CT is a useful tool in preoperative diagnosis of Amyand's hernia but this is not a routine investigation in children presented with incarcerated or strangulated hernias [14]. The pathophysiology of Amyand's hernia is still unclear but the acute inflammation of the appendix within the hernia sac may be due to compression by the sharp edge of the internal ring that results in pressure necrosis, bacterial spread, and appendicitis [2, 15]. There are controversies in the literature about dealing with normal looking appendix within the hernia sac when incidentally found during surgery. Many authors recommended repair of the hernia without appendectomy in this condition [2, 8, 16] but others preferred appendectomy in all patients [17, 18].

In this study, we presented our experience in the management of 12 pediatric patients presented with AH over 10 years at tertiary referral hospital and highlight the difference in dealing with normally looking appendix between pediatric and adult patients.

Methods

A retrospective analysis of the charts of all patients operated for congenital inguinal hernia presented at Tanta university hospitals and its affiliated hospitals, Egypt, in the period from December 2009 to December 2019. Patient's demographics (age, sex), clinical presentation (history, clinical examination), preoperative investigations (routine investigations and imaging studies), surgical notes, management strategy, and postoperative results were recorded and analyzed.

Results

The charts of 2500 cases of congenital inguinal hernia were reviewed. AH was found in 12 (0.48%) patients presented by congenital inguinal hernia. Ten patients were boys, and two patients were girls. The median age at the operation was 7 months (range 15 days to 5 years). The clinical presentation was incarcerated inguinal swellings in all patients. Laboratory findings were normal in all patients except one of them who had elevation in the total leucocytic count and CRP found during surgery to be inflamed appendix. Preoperative inguinoscrotal ultrasonography was done in 3 patients only and revealed an incarcerated tubular structure in the hernia sac in 2 patients and incarcerated bowel in one patient which was discovered during surgery to be a cecum and appendix. Emergency surgery was done in all patients. Eleven patients were operated through transverse inguinal incision at the lower abdominal crease, and a normally looking appendix alone was found in the hernia sac in 9 patients which were reduced easily, and herniotomy was done (Fig. 1a–c). The hernia sac was found to contain

a sliding cecum with a normal looking appendix in one patient which was reduced with some difficulties, and herniotomy was done (Fig. 2). Inflamed appendix was found in the one patient, and appendectomy was done which facilitates reduction and closure of the hernia sac (Fig. 3). One patient was operated using laparoscopy; the appendix was found normally looking and adherent within the hernia sac; reduction of the appendix was done by external scrotal compression; and the adhesion was dissected using laparoscopic scissor and monopolar diathermy, then the sac was closed using burse string suture at the internal ring (Fig. 4) (Table 1). All patients were discharged at the same day of the operation, and the postoperative course was uneventful in all cases. The patients were followed in the outpatient clinic at 2 weeks, 1 month, and every 3 months. No recurrence was detected in any patient in the follow-up period which extended for 9 months after the operation.

Discussion

Amyand's hernia is a rare condition and defined as presence of vermiform appendix within inguinal hernia sac. Regarding the pathophysiology of AH, some reports explained that the presence of fibrous connection between the appendix and the testis acts as a guidance to the passage of the appendix with the aid of patent processus vaginalis into inguinal hernia. So, AH was reported in neonates and premature twins [12, 19–21]. Other reports explained AH by the repeated microtrauma, adhesions, and recurrent acute inflammation which occurred due to migration of the appendix within the hernia sac [22, 23].

Clinically, the preoperative diagnosis of AH is difficult, and most cases were presented 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 AH. The general manifestations like fever, vomiting, and abdominal distention depend on the condition of the vermiform appendix whether inflamed or perforated, and the clinical image may be obscured due to the narrow neck of the sac of the herniation which limits the spread of inflammation and peritoneal irrigation [24]. The differential diagnosis of AH also included torsion of the testis and epididymo-orchitis [25].

Inguinoscrotal ultrasonography and CT scan are important tools in predicting this condition [6]. In this study, all patients were presented as right inguinal and inguinoscrotal swellings. Clinical examinations revealed incarceration in all patients without the symptoms and signs of intestinal obstruction. One patient

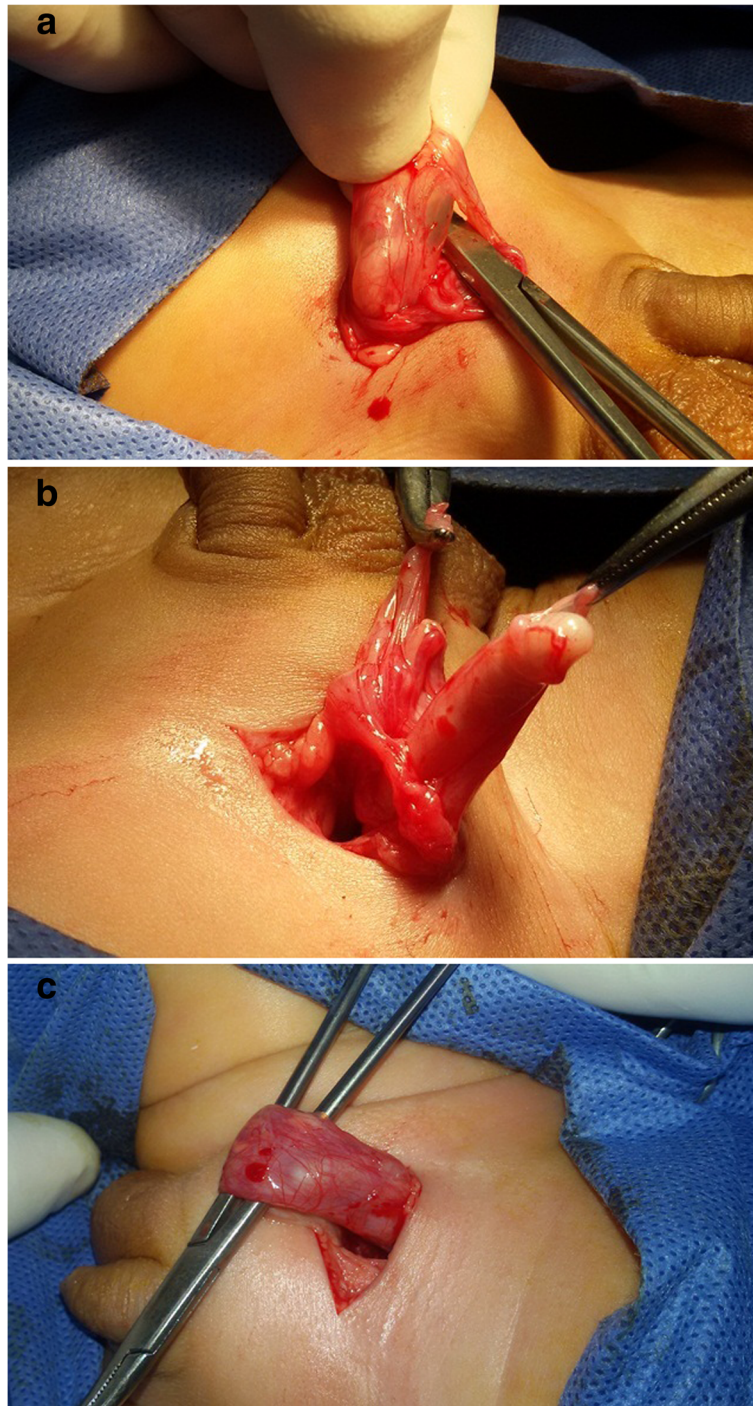


Fig. 1 a-c AH containing normally looking appendix

was suspected clinically to be AH with inflamed appendix because of the general and local manifestation of appendicitis, and the diagnosis was confirmed by laboratory findings and inguinoscrotal ultrasonography. During surgery, acute appendicitis was found,

and the appendix was too long and it was descended to the scrotum, and appendectomy was done. Priego et al. reported no cases with acute inflammation of the appendix in their series which include 6 adult patients with AH [17]. Kaymakci et al. had one case of



Fig. 2 AH containing sliding cecum and appendix

appendicitis in their series which included 30 pediatric patients [26]. Cankorkmaz et al. had 4 cases with acute appendicitis in their series which included 12 pediatric patients [8]. Okur et al. reported also 4 patients with inflamed appendix in their series which included 21 pediatric patients [16]. Milburn and Youngson reported a 10-day-old boy with AH presented with acute hemiscrotum [27]. Livaditi et al. reported two premature neonates with AH, and one of them was found to have perforated appendicitis [28].

Losanoff and Basson described a classification system in 2007 clarifying the operative approach, the methods of dealing with the appendix, and the type of the hernia repair in adults (Table 2) [29], but there is still lacking of consensus about dealing with the

normal looking appendix in pediatric patients presented with AH.

In cases of AH with appendicitis, it is accepted that appendectomy should be performed [26, 30] but the controversies about AH with normal looking appendix is still present. Many authors believed that the normally looking appendix which was incidentally discovered during surgery without any signs of inflammation should not be removed, and prophylactic appendectomy is not necessary [2, 8, 16]. They concluded that, the unnecessary appendectomy may add an operative risk and leads to dissemination of the infection. Also, dissection at the base of the appendix may need extension of the incision and so weakness of the tissues and hence further recurrence;



Fig. 3 AH containing inflamed appendix

finally, the appendix may be used in later life in other operations like urinary diversion. We support this opinion in this study as reduction of the normally looking appendix, and repair of the hernia was done in 11 patients, and there were no post-operative complications or recurrences. Other authors choose to make appendectomy to all patients presented with AH. They explained that opinion by the high chance of the appendix to herniate in young patients leading to recurrence may lead to later appendicitis [17, 18].

Advances of minimal invasive surgery in the last 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, 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 deferens and testicular vessels to avoid injury of

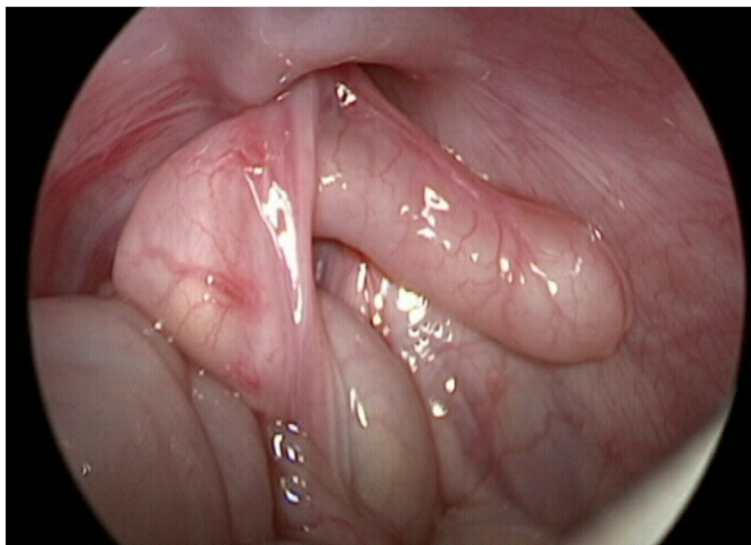


Fig. 4 Laparoscopic view of AH after reduction of the appendix

Table 1 Patients characteristics

Patients characteristics	Number
Male	10
Female	2
Site	
1-Unilateral CIH	9
2-Bilateral CIH	3
Preoperative ultrasound	3
1-Incarcerated appendix	2
2-Incarcerated bowel	1
Procedure	
1-Traditional open approach	11
2-Laparoscopic approach	1
Findings	
1-Normal looking appendix alone	10
2-Sliding cecum and normal appendix	1
3-Inflamed appendix	1

both structures [31]. Chan et al. reported advantages of the laparoscopic approach over an open approach with regards to postoperative pain, recovery, and wound cosmesis [32]. Tycast et al. reported a 12-year-old boy with AH who was managed by laparoscopic appendectomy, and the procedure was completed by open repair of inguinal hernia [33]. Rahman et al. reported a 10-week-old boy presented with AH managed laparoscopically [31]. In this series, there was only one case done laparoscopically where there was adhesion of the appendix within the hernia sac which was easily dissected and reduced under vision aided by external scrotal compression. The appendix was looking normal and so appendectomy was not done.

The main limitations of this study are the small sample size which could be attributed to the rarity of the condition and the study format as a case series with lack of the control group.

Table 2 Losanoff–Basson classification of Amyand's hernia (AH) and their management [28]

Types of AH	Features	Surgical management
Type 1	Normal appendix within the inguinal hernia	Reduction of appendix or appendectomy and mesh hernioplasty
Type 2	Acute appendicitis with no abdominal sepsis	Appendectomy through the hernia and sutured hernioplasty
Type 3	Acute appendicitis with abdominal sepsis	Appendectomy through laparotomy with sutured hernioplasty
Type 4	Acute appendicitis associated with related or unrelated abdominal pathology	Appendectomy through hernia or laparotomy plus diagnostic workup

Conclusion

Amyand's hernia is a rare condition and is always misdiagnosed as strangulated hernia. It should be suspected in cases with long history of incarcerated congenital hernia without clinical manifestation of intestinal obstruction. Careful examination of the appendix must be considered, and routine appendectomy is not mandatory for a normal looking appendix in pediatric patients.

Abbreviations

AH: Amyand's hernia

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Authors' contributions

HAA gave the idea and collected the patients' data and analyzed them. MAS wrote the paper with revision and the corresponding author. MAM put the study design and followed the patients postoperatively. All authors have read and approved the final version of the manuscript.

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Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request. Available upon request

Ethics approval and consent to participate

This study was approved by the Review Board of the Department of General Surgery, Faculty of Medicine, Tanta University, Egypt, on 15 June 2019, but the consent to participate was not applicable as it is a retrospective study. The ethical committee reference number: 2938/12/14.

Consent for publication

Not applicable as it is a retrospective study.

Competing interests

All authors have no conflicts of interest or financial ties to disclose. Contents have not published elsewhere, and the paper is not being submitted elsewhere.

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