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Case Report

Our experience with Amyand's hernia

Brajesh Gupta, Ghanshyam Hatwar*, Charu Tiwari, Shweta Gupta

Department of Surgery, Government Medical College and Hospital, Nagpur, Maharashtra, India

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***Correspondence:**
Dr. Ghanshyam Hatwar,
E-mail: gmhatwar@gmail.com

ABSTRACT

Amyand's hernia is a rare type of inguinal hernia with appendix as its content, which may be normal or pathological. We present our experience with Amyand's hernia in 6 patients. A retrospective review of 6 cases with Amyand's hernia that were managed over a 2-year period was done by reviewing the hospital records. The variables assessed and analyzed were demographic details, clinical presentation, management and follow-up. There were 4 children and 2 adults; male and female ratio being 5:1. The mean age of four children was 1.5 years; the two adults were 72 and 54 years of age. The duration of symptoms of inguinal hernia ranged from 1 month to one year; mean duration of irreducibility was 5.33 days (range: 2 days to 15 days). Five patients had symptoms of obstruction. All 5 patients with obstruction underwent emergency surgery. The sixth patient was posted for elective hernioplasty the next day. Appendix was inflamed only in one patient. The post-operative course was uneventful in all patients. The mean duration of hospital stay was 3.33 days. Only one patient had surgical site infection. Amyand's hernia, thus presents with features of incarcerated right hernia. Pre-operative diagnosis is difficult. Intra-operatively, the surgical procedure needs to be individualized depending upon the pathology.

Keywords: Amyand's hernia, Inguinal, Appendix

INTRODUCTION

Inguinal hernia is one of the common surgical conditions encountered in surgical practice. Inguinal hernia is broadly divided into direct and indirect inguinal hernias. An inguinal hernia usually will contain either small or large bowel, or only omentum. Less common and often diagnosed intra-operatively, contents can be unexpected such as in the case of Meckel's diverticulum (Littre's hernia), appendix [Amyand's hernia (AH)], a segment of the bowel's circumference (Richter's hernia) or even a portion of the bladder (sliding hernia).[1]

An AH is characterised by whole or part of appendix as content of an inguinal hernia; the appendix itself may be normal, inflamed or exhibiting any other pathology.[2] The reported incidence of AH is as low as 1% and complication with acute appendicitis is reported in only 0.08–0.13% of cases.[3] A perforated appendix in an inguinal hernia sac is even rarer.

The signs and symptoms of AH can be variable, depending on the appendiceal pathology within. However, the initial clinical presentation of inguinal hernia can mask a variety of other pathologies as acute appendicitis, appendicular perforation appendicular abscess or even malignancy.[3]

Because of the rarity of this type of hernia and only scattered case reports published in literature, the pathophysiology of this condition is enigmatic and a standard management approach has not yet been established.[2] We describe our experience with AH over a 2-year period at our institute. A brief review and discussion of literature of this rare hernia follows.

CASE REPORT

A total of 6 patients with AH were managed over a period of one year. There were 4 children and 2 adults; male and female ratio being 5:1. The details are summarized in Table 1.

Clinical presentation

The mean age of four children was 1.5 years; the two adults were 72 and 54 years of age. All 6 patients presented with symptoms of right inguinal hernia which became irreducible (Figure 1). The duration of symptoms of inguinal hernia ranged from 1 month to one year; mean duration of irreducibility was 5.33 days (range: 2 days to 15 days). Five patients had symptoms of obstruction bilious vomiting, abdominal distention and constipation of mean duration of 1.4 days. The sixth patient had no associated symptom other than irreducibility of hernia.

Figure 1: Clinical image of right sided irreducible hernia in a 1 year child.

Table 1: Clinical presentation and management of 6 patients with Amyand's hernia.

S. No.	Age (in years)	Sex	Clinical presentation	Intra-operative finding (content)	Surgery
1	2	M	Rt irreducible hernia with bilious vomiting, abdominal distention and constipation	Caecum with normal appendix and ileocaecal junction	Herniotomy
2	2	M	Rt irreducible hernia with bilious vomiting, abdominal distention and constipation	Caecum with inflammed appendix, ileocaecal junction, terminal ileum and omentum	Herniotomy and appendicectomy
3	1	F	Rt irreducible hernia with bilious vomiting, abdominal distention and constipation	Caecum with normal appendix, ileocaecal junction, terminal ileum and omentum	Herniotomy
4	1	M	Rt irreducible hernia with bilious vomiting, abdominal distention and constipation	Caecum with normal appendix, ileocaecal junction, terminal ileum and omentum	Herniotomy
5	54	M	Rt irreducible hernia with bilious vomiting, abdominal distention and constipation	Oedematous caecum, normal appendix and omentum	Herniorraphy and partial omentectomy
6	72	M	Rt irreducible hernia only	Caecum, normal appendix and omentum	Hernioplasty with prolene mesh

Tachycardia was present in 5 patients. Right-sided irreducible hernia was present in all 6 patients. Five patients had tenderness and local rise of temperature with redness of overlying skin. Abdomen was distended and tender in right iliac fossa in 5 patients. Exaggerated bowel sounds were present in 2 patients while they were absent in 3 patients. The sixth patient with no abdominal complaints had no per abdominal signs.

Management and follow-up

Leucocyte counts were raised in all patients. Abdominal erect X-ray showed right inguinal hernia with features of obstruction– distention and presence of air-fluid levels in five patients (Figure 2). The sixth patient had no obstruction on X-ray. Ultrasonography confirmed right irreducible hernia and revealed normal vascularity of herniated bowel in all cases.

Figure 2: X-ray abdomen of a patient with Amyand's hernia.

All 5 patients with obstruction were taken for emergency surgery. The sixth patient was posted for elective hernioplasty the next day. The details of intra-operative findings and surgery done have been summarized in Table 1. Appendix was inflamed only in one patient who underwent appendicectomy (Figure 3). Rest all patients had normal appendix; hence appendicectomy was not done (Figure 4). The post-operative course was uneventful in all patients. Oral feeds were started the next day. The mean duration of hospital stay was 3.33 days. Only one patient had surgical site infection. There was no mortality. All patients are doing well on follow-up.

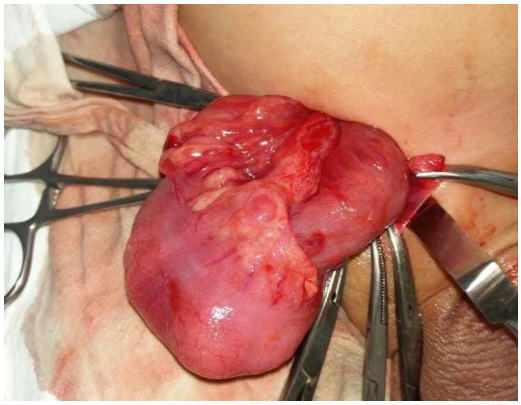

Figure 3: Intra-operative image showing inflamed appendix and caecum in the hernia sac of a child.

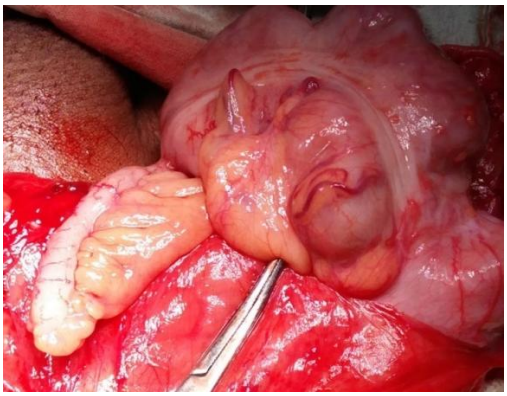

Figure 4: Intra-operative image showing normal appendix and caecum in the hernia sac of an adult.

DISCUSSION

An AH is an inguinal hernia which contains whole or part of appendix in the sac.[2] This is a rarely encountered entity and accounts for approximately 1% of all inguinal hernias, with some reports stating incidence to be as low as 0.04 to 0.06%.[2] The incidence becomes even lower when appendix is pathological, i.e., inflamed (0.07-0.13%) or complicated by perforation and abscess formation (0.1%).[1] Appendix has been known to protrude into the inguinal AH and femoral (De Garengeot's hernia) hernia sacs; and less commonly, into obturator hernia, umbilical hernia, Spigelian hernia, laparoscopic port site hernia, drain site hernia, incisional hernia and diaphragmatic hernia.[4]

AH was first described by Amyand; approximately 228 such cases have been documented since then.[5] Due to its rarity, paucity of literature and clinical presentation indistinguishable from that of an incarcerated hernia, the etiopathogenesis of this condition is still unclear and management guidelines are also lacking.

In terms of the cases reported in the literature thus far, the age of patients who have had an AH varies greatly, but is commonly found in males and in paediatric population (due to the presence of patent processus vaginilis).[2] Our study also corroborates with literature in terms of age and sex.

The pathophysiology is still debated regarding the cause of appendiceal herniation and development of appendicitis within the sac; one explanation being a previously obstructed appendix due to a faecolith or lymphoid hyperplasia, which becomes entrapped and inflamed within hernia sac due the original obstruction rather than the entrapment; however, there are only few reports of Amyand's appendicitis associated with fecoliths or villous adenoma.[3,5,6] There are theories stating that it is secondary to a patent processus vaginalis or perhaps related to the presence of a fibrous band between the hernia sac and testes.[2]

Other postulated mechanisms include adhesions causing irreducibility of the hernia and compression of the appendix in the external ring resulting from increases in intra-abdominal pressure.[7] A systematic review by Michalinos et al reported that most authors believe that the appendicitis develops due to external compression from muscle contraction and sudden increases in intra-abdominal pressure causing ischemia and subsequent inflammation.[8]

AH is mostly an indirect inguinal hernia; but it may rarely appear as a direct inguinal hernia.[4] Generally, only appendix is found inside the hernia sac in paediatric age group; but cecum or other small bowel loops may be found in adults.[4] An AH is more frequently found on the right, possibly due to the anatomical position of the appendix as well as the fact that right inguinal hernias tend to be more common. A few cases of left sided AH have been reported and in those cases, it is important to consider an associated condition which would enable such a variation, including situs inversus, malrotation of gut or increased mobility of the caecum.[9] Few cases of appendicular malignancy have been reported in AH.[10-12] In this study, all hernias were right sided and appendix was inflamed only in one patient.

The most common presentation is that of a painful scrotal inguinoscrotal mass, usually representative of an incarcerated or strangulated hernia; however, it can also present with small bowel obstruction, scrotal abscess,

epididymitis or even as an acute scrotum.[2] It has also been described in cadavers, which suggests that it can be asymptomatic.[2] Variable presentations relate with the status of the appendix as well as whether there is caecal involvement within the hernia.[2] All our patients presented with irreducible right inguinal hernia with features of obstruction seen in 5 of them.

Since the standard of care for such patients is emergent surgical repair, it follows that AH are almost exclusively diagnosed intraoperatively.[3] Pre-operative diagnosis is uncommon. Preoperative diagnosis of an AH is infrequent and has only been reported in a limited amount of cases. In most cases, AH is diagnosed intra-operatively.[13] Sharma et al reported 18 patients with AH, while no patient was diagnosed pre-operatively.[14] Cankormaz et al reported 12 neonates and infants with AH, and pre-operative diagnosis was made in only 1 case.[15] Weber et al reported a retrospective study of 60 AH cases, treated over 12 years, out of which pre-operative diagnosis was made in only one patient.[16]

Vehbi et al used ultrasonography and confirmatory computed tomography (CT) scan to successfully diagnose an AH in a 49-year-old female whereas a few others have incidentally, yet successfully identified AH based on preoperative CT scans.[17-20] However, most surgeons do not recommend imaging examination for preoperative evaluation, in order to proceed with surgical repair of an inguinal hernia, especially when there is indication of prompt therapeutic intervention.

In order to help guide and improve treatment of AH, Losanoff and Basson et al devised a widely accepted classification and management scheme that divides AH into four subtypes.[21]

Type 1

Normal appendix managed by reduction or appendectomy (depending on age), mesh hernioplasty.

Type 2

Acute appendicitis localized in the sac managed by reduction or appendectomy (depending on age), mesh hernioplasty.

Type 3

Acute appendicitis, peritonitis managed by appendectomy through laparotomy, endogenous repair.

Type 4

Acute appendicitis, other abdominal pathology managed by appendectomy, diagnostic workup and other procedures as appropriate.

A modification of the Losanoff and Basson classification of AH, also known as Rikki modification by Singal et al has also been propsed wherein a fifth type of AH is added on the latter classification, referred as an incisional hernia through which the vermiform appendix protrudes. This type is further divided into three subtypes, 5a, 5b and 5c.[22]

After intra-operative diagnosis, the management is to be individualized based on the pathological features present.[3] The most commonly encountered pathological features are normal appendix, acute appendicitis, perforated appendicitis and periappendicular abscess.[4] However, a very limited number of cases of an appendiceal tumoral lesion inside the AH sac has been reported in the literature.[4]

When there is appendicitis or a perforated appendix, it is widely accepted that an appendectomy should be performed in addition to hernia repair, avoiding the use of mesh due to the danger of infection.[3] If a non-inflamed appendix is found, appendicectomy is usually not recommended; however, some advocate for a prophylactic appendectomy on the grounds that it is prone to re-herniate and may cause future appendicitis.[3]

For appendicular tumours, right hemicolectomy at the same surgery, or after the pathology report, has been reported as the most appropriate approach for appendicular adeno-carcinoma, irrespective of their location and size.[4] Appendicular mucocele should be resected with clean borders, as there may be a potential for malignancy.[4]

According to Losanoff's classification, if an appendectomy is performed then mesh should not be placed to avoid the risk of infection. However, Kose et al argues that in the situation with a healthy appearing incarcerated appendix with no signs of inflammation an appendectomy can be performed along with mesh hernioplasty.[23]

The use of mesh to repair the hernia has also been a topic of controversy. Some see it as a contraindication due to the increased chances of having an inflammatory response from the contaminated abdominal wall and the synthetic prosthesis.[24,25] Sharma et al argue that it is safe to retain a normal appendix and to use mesh for the hernia repair.[14]

The importance of recognising this condition lies in the fact that a variable, yet significant, mortality has been associated with it (5.5–30%), mostly due to peritonitis and sepsis.[26] Complications from an inflamed or perforated appendix contained in an AH can extend to involve the right testicle and associated structures, form intra-abdominal abscesses or even cause necrotising fasciitis of the inguinal region or anterior abdominal wall.[1]

CONCLUSION

An AH poses a diagnostic challenge, as its identification is often incidental, on imaging or intra-operatively. An AH can present with a range of signs and symptoms, depending on the appendiceal pathology within. More specifically, an AH can contain a normal, asymptomatic appendix or an abscess formed secondary to perforation of an acutely inflamed appendix. Because of the rarity of this type of hernia, a standard management approach has not been established and the management needs to be individualized.

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