CASE REPORT

Amyand's Hernia Study of four cases and literature review

*Syed M. Ali, Kamran A. Malik, Hani Al-Qadhi

الملخص: فتق امياند هو عبارة عن احتواء الزائدة الدودية بكيس الفتق الآربي. يعتبر فتق امياند من الحالات النادرة التي لا تكتشف غالبا إلا أثناء التدخل الجراحي للفتق الآربي. قد تكون الزائدة الدودية بكيس الفتق بحالتها الطبيعية أو تكون بحالة ملتهبة مع المضاعفات المحتملة. ندرج في هذا التقرير وصفا لأربع حالات من فتق امياند بمستشفى جامعه السلطان قابوس، خضعت هذه الحالات للجراحة باستئصال الزائدة الدودية مع إصلاح الفتق بشبكة طبية في ثلاث حالات، أما الحالة الرابعة فقد قمنا بإصلاح الفتق بدولين.

مفتاح الكلمات: الفتق الآربي، زائدة دودية داخل فتق، التهاب الزائدة الدودية الحاد، تقرير حالة، عُمان.

ABSTRACT: The presence of the appendix in an inguinal hernial sac is described as Amyand's hernia. It is a rare entity which presents mostly at the exploration of the inguinal canal. The appendix may be apparently normal or have all the features of acute appendicitis with its possible complications. We report four cases of Amyand's hernia which were treated at Sultan Qaboos University Hospital, Oman. All patients underwent appendectomy. In three cases, the inguinal hernia were repaired with Vipro mesh while, in the remaining case, a darning repair was done with Prolene sutures.

Keywords: Inguinal hernia; Appendix in hernia; Acute appendicitis; Case report; Oman.

HE CONTENTS OF INGUINAL HERNIA SACS differ from case to case. Various structures contained therein have been described, but the presence of the appendix in an inguinal hernia sac is rare. This anomaly was first described by Claudius Amyand in an 11-year-old boy who underwent a successful appendectomy in 1735.¹ The incidence of appendicitis within an inguinal hernia is estimated at 0.07–0.13 %.² The eponym Amyand's hernia was first coined by Creese in 1953, then by Hiatt and Hiatt in 1988, followed by Hutchinson in 1993.^{3–6}

We report four cases of Amyand's hernia operated upon in the Department of Surgery at Sultan Qaboos University Hospital, Oman, in the period 2007 to 2011. Patient follow-up ranged from one month to three years with no surgical site infection seen in the immediate post-operative period, or recurrence of the hernia. The purpose of this report is to create general awareness among surgeons who might be dealing with this hernia surgery, as they may encounter unexpected intraoperative findings such as Amyand's hernia. It is important to be aware of all clinical possibilities and appropriate management techniques.

Case One

A 64-year-old male, who had had a left-sided reducible inguinal hernia for the previous 5 years, presented with a 2-day-history of fever, pain, vomiting, and irreducibility of the hernia. He had noticed a recent increase in the swelling size. On examination, he was found to be dehydrated with a temperature of 38° C and a heart rate of 104 beats per minute (bpm).

He had bilateral pedal oedema and bibasilar crepitations in his chest. His abdomen was distended with exaggerated bowel sounds. There was a 30 x 15 cm pear-shaped left inguino-scrotal

Department of Surgery, Sultan Qaboos University Hospital, Muscat, Oman *Corresponding Author e-mail: ali_2051@hotmail.com, and alismc2051@gmail.com



Figure 1: Amyand's hernia in patient 2: (A) right inguinal hernia sac, (B) appendix.

swelling extending up to the base of the scrotum. The lump was tense, tender, and irreducible. Both testes were palpable in the scrotum. After adequate resuscitation he was taken to surgery under general anaesthesia.

The left inguinal canal was explored, and a strangulated irreducible indirect inguinal hernia was found, with constriction at the external ring. About 1.2 litres of an amber-coloured fluid was aspirated upon opening the sac, and the gangrenous caecum and appendix were removed. The small bowel loops were dusky initially, but became pink upon release of the constriction The contents of the sac were pushed inside and a lower midline laparotomy was performed. A gangrenous floppy mobile caecum was seen, along with a gangrenous appendix which had herniated through the deep ring on the left side. There was no situs inversus or malrotation of the gut. The rest of the viscera were normal. A limited right hemicolectomy was done with ileocolic anastomosis. Before closing the abdomen, the hernia was repaired by darning with size 0 Prolene (Ethicon, Inc., Manlo Park, California, USA). The patient was treated with broad spectrum

antibiotics and had an uneventful recovery. The histopathology was consistent with a gangrenous caecum and appendix. After one year, the patient was well with no recurrence of the hernia.

Case Two

A 19-year-old male was admitted for right inguinal hernia repair. He had had the hernia for six months. It was increasing in size, especially during walking, but was reducible upon lying down. Examination showed swelling in the right inguinal region, which was non-tender, partially reducible, and had a positive cough impulse. The patient underwent inguinal hernia repair under general anaesthesia. Per-operatively, he was found to have an indirect sac containing a congested appendix with palpable fecolith. An appendectomy was performed and repair of the hernia was accomplished with Vipro mesh (Ethicon, Inc., Manlo Park, California, USA). The patient had an unremarkable recovery and was discharged three days after the surgery. He was seen for follow-up after one month; the incision site was uninfected. The histopathology result showed a

Classification	Description	Surgical management
Type I	Normal appendix in inguinal hernia	Hernia reduction, mesh repair; appendectomy in young patients
Type II	Acute appendicitis within an inguinal hernia and no abdominal sepsis	Appendectomy through hernia; primary repair of hernia; no mesh
Type III	Acute appendicitis within an inguinal hernia or the abdominal wall, or peritoneal sepsis	Laparotomy; appendectomy; primary repair of hernia; no mesh
Type IV	Acute appendicitis within an inguinal hernia with related or unrelated abdominal pathology	Manage as hernias type I–III; investigate or treat second pathology as appropriate

Table: Classification of Amyand's hernias8

congested appendix with lymphoid hyperplasia.

inflammation. He was doing well three years after surgery.

Case Three

A 75-year-old male presented with a 2-month history of swelling in the right inguinal region, which had increased recently causing discomfort but no significant pain. It was reducible with incomplete swelling, but the patient had a positive cough impulse. On exploration of the inguinal canal, the appendix was discovered to be adherent to an indirect hernia sac. The appendix was normal looking, but was removed after dealing with the adhesions. The histopathology revealed an inflamed appendix. The posterior wall was repaired with Vipro mesh (Ethicon, Inc., Manlo Park, California, USA) and the patient was discharged two days after surgery. He was seen for follow-up one month after surgery and there was no infection at the surgical site. On his last visit to the surgical clinic, two years after surgery, no recurrence was found.

Case Four

A 26-year-old male was admitted for an elective repair of a right-sided inguinal hernia. He had had reducible swelling for the previous two years. The hernia had recently become partially irreducible, but the patient was without pain or any other symptoms other than a positive cough impulse. Per-operatively, it was noted that he had an indirect inguinal hernia, and a caecum and appendix which were not inflamed on gross appearance. An appendectomy was performed along with repair of the hernia with Vipro mesh (Ethicon, Inc., Manlo Park, California, USA). He recovered without any complications. Histopathology of the appendix was consistent with lymphoid hyperplasia without any Discussion

A hernia is the protrusion of the viscus or a part of the viscus through the wall of its containing cavity. By far the most commonly encountered hernia is in the inguinal region which also normally contains bowels, or omentum. Among the unusual contents are the bladder, Meckle's diverticulum (known as Littre's hernia), or a portion of the circumference of the intestine (called Richter's hernia), but Amyand's hernia is relatively unknown despite being first reported in 1735 by Claudius Amyand.¹

The term Amyand's hernia is used to refer to a hernial sac containing an inflamed or non-inflamed appendix in an irreducible inguinal hernia.⁷ Losanoff and Basson suggested a distinct classification to improve the management of Amyand's hernias.⁸

The incidence of a normal appendix being found inside an inguinal hernia sac is about 1%; however, only 0.1% of these cases have appendicitis.⁹ Solecki *et al.* observed that acute appendicitis was found in 0.62% of all groin hernia sacs.^{10,11} In most of the patients who present with a right-sided Amyand's hernia, its location can be explained by the normal anatomical position of the appendix; also, rightsided inguinal hernias are more common. In this study, three patients had right-sided hernias. However, left-sided Amyand's hernias have also been described in the literature and may be associated with *situs inversus*, malrotation of the gut, or mobile caecum, as was found in one of our cases.^{6,12}

The pathophysiology of Amyand's hernia is unknown. Weber *et al.* proposed that due to herniation the appendix can become more vulnerable to micro-trauma, causing adherence to the hernia sac due to fibrosis.¹³ This hypothesis that inflammatory swelling may lead to incarceration, subsequent impaired blood supply, and bacterial overgrowth was supported by Abu Dalu, Barut, and House.^{14–16}Muscle contractions and changes in abdominal pressure can cause compression of the appendix, resulting in reduced blood supply and secondary inflammation.¹¹

Diagnosing Amyand's hernia pre-operatively is not straight forward. In the majority of cases, it is diagnosed when the hernia sac is opened, as most patients undergo emergency surgery. Although a preoperative computed tomography (CT) scan of the abdomen can be helpful in diagnosing the condition, it is not routinely employed in such cases. If the diagnosis is established by CT, it is possible to treat Amyand's hernia laparoscopically.¹⁷

The recommended treatment is appendectomy with primary hernia repair. Use of synthetic mesh is avoided in the repair of contaminated abdominal defects because prosthetic material can increase the inflammatory response and result in wound infection and a rare but possible complication of appendiceal stump fistula.^{18–20} Preigo *et al.* carried out appendectomies in six patients, using mesh in three. One patient developed a wound infection after being treated with mesh.²¹ Bailey reported a wound infection rate of 3% in hospital that went up to 9% in community surveillance.²² However, Saggar *et al.* reported endoscopic total extraperitoneal repair with mesh in a right-sided incarcerated inguinal hernia without any complications.²³

We performed appendectomies in all of our cases and repaired the hernia with mesh in three of our patients. In Case 1, we decided not to use mesh due to the presence of a gangrenous appendicitis. This guarded the hernia from a possible future extension of inflammation into the mesh. However, we did repair the hernia with mesh in cases 2 to 4 as the appendices were mildly inflamed with no purulent fluid in the hernial sac. The follow-up period in our patients ranged from one month to three years with no surgical site infection seen during the immediate post-operative period. No recurrence of the hernias was found.

Conclusion

Amyand's hernia is a rare clinical entity that is

difficult to diagnose pre-operatively. The presence of an inflamed or gangrenous appendix increases the rate of complication, particularly increasing the rate of wound infection. Diagnosis is usually made at the time of surgery, which is usually indicated in all incarcerated hernias. Consequently, our recommendation is that the decision to perform an appendectomy and/or to use mesh to repair hernias should always be individualised.

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