

Amyand's Hernia: A Ten-year Experience With 6 Cases

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Abstract

Purpose: Amyand's hernia is a rare hernia defined as an inguinal hernia that contains the appendix within the hernia sac. Current treatment of Amyand's hernia remains controversial. Our study retrospectively reviewed 6 cases of Amyand's hernia, aiming to provide a reference for the surgical treatment of Amyand's hernia.

Methods: Six patients diagnosed with Amyand's hernia from September 2010 to May 2020 were retrospectively enrolled in our study. We summarized clinical data of six patients including the chief complaint, physical examinations, laboratory examinations, imaging examinations, surgical methods, and postoperative treatments and outcomes.

Results: The diagnosis of six cases with Amyand's hernia was made during surgery. Two patients had normal appendixes whereas the remaining four patients had inflamed appendixes. Two patients with normal appendix received tension-free mesh repair through the inguinal incision. Among those with inflamed or perforated appendixes, two received mesh repair and the other two did not. The discharge time after surgery of six patients was 9.8 ± 6.1 days. One patient suffered from an incision infection. No additional postoperative complications were detected.

Conclusions: Computed tomography and ultrasonography are helpful but limited in the definite diagnosis of Amyand's hernia. The presence of a normal appendix does not require to be resected, but appendectomy is necessary if the appendix is inflamed. The treatment of Amyand's hernia should be tailored based on the patient's condition and the type of Amyand's hernia.

Introduction

Amyand's hernia is defined as an inguinal hernia that contains the appendix within the hernia sac [1]. In 1735, C. Amyand first described an 11-year-old boy with an incarcerated inguinal hernia containing a perforated appendix [2]. Subsequently, this type of hernia was named Amyand's hernia, which was rarely encountered in clinical practice. Amyand's hernia occurs in only 1% (0.19–1.7%) of all inguinal hernia cases [3, 4, 5, 6]. Moreover, Amyand's hernia is classified into four subtypes regarding the clinical symptoms and the situation of the appendix [8]. The appendix within the hernia can be either normal or inflamed, in which 0.13% of cases have inflamed appendix. Notably, the perforation of the appendix could lead to a dramatic increase in the mortality rate (15–30%) due to severe abdominal sepsis [3, 4, 5, 7]. Commonly, the diagnosis of Amyand's hernia was made intraoperatively and few cases could be diagnosed before surgery. Besides, current management of Amyand's hernia remains controversial since different strategies should be tailored to different individuals. In this study, we reported six cases of Amyand's hernia and detailed different managements, aiming to provide a reference for the surgical treatment of Amyand's hernia.

Patients And Methods

The study protocol, conform to the ethical guidelines of the 1975 Declaration of Helsinki, was approved by the Institutional Ethical Review Committee of The Dalian Municipal Central Hospital Of Dalian Medical University. Written informed consents were obtained from all the participants. Six patients diagnosed with Amyand's hernia from September 2010 to May 2020 were retrospectively enrolled in our study. The following data were collected for further analysis: age, sex, chief complaint, clinical manifestations, white blood cell (WBC) and C-reactive protein (CRP) level at admission, diagnostic imaging, surgery, and postoperative outcome. The level of WBC and CRP of six patients were matched with the type of Amyand's hernia which was diagnosed intraoperatively. WBC higher than $11 \times 10^9/L$ and CRP higher than 10 mg/L were defined as elevated, which indicated the existence of inflammation. The primary endpoint was 30-day hospital mortality. Secondary endpoints were postoperative surgical wound infection and hospital length after surgery.

Results

Characteristics of six patients

A total of six patients with Amyand's hernia were admitted to the hospital. All of them were males with the age of 64.0 ± 17.6 years old (range: 30–81 years old) (Table 1). One patient (16.7%) was admitted to the hospital because of right lower abdominal pain, and five patients (83.3%) were admitted because of a mass in their inguinal area and right lower abdominal pain.

Table 1

Baseline characteristics of six patients

Case No.	Age (years old)	Sex	Clinical presentation	Diagnostic imaging	WBC and/or CRP
1	77	Male	Inguinal hernia associated with right lower quadrant abdominal pain	Abdominal CT	Elevated
2	64	Male	Inguinal hernia associated with right lower quadrant abdominal pain	Abdominal CT	Elevated
3	56	Male	Inguinal hernia associated with right lower quadrant abdominal pain	Ultrasonography	Normal
4	30	Male	Right lower abdominal pain	Ultrasonography	Elevated
5	77	Male	Inguinal hernia associated with right lower quadrant abdominal pain	Abdominal CT	Elevated
6	71	Male	Inguinal hernia associated with right lower quadrant abdominal pain	Abdominal CT	Normal

Preoperative examinations

Six patients underwent computed tomography (CT) or ultrasonography examination before the operation. Four patients (Case 1, 2, 5, and 6) were diagnosed with appendicitis and inguinal hernia by abdominal CT. The appendix had a similar density compared with the surrounding hernia on CT images (Fig. 1). Case 3 was diagnosed with the inguinal hernia by ultrasonography whereas an enlarged appendix lumen and a right inguinal hernia were discovered by ultrasonography in Case 4. In laboratory examinations, WBC and CRP were helpful for distinguishing the inflamed appendix from the normal one. WBC and CRP were normal in two patients (Case 3 and 6) who had non-inflamed appendixes; in contrast, the other four patients (Case 1, 2, 4, and 5) with inflamed or perforated appendix exhibited elevated WBC and/or CRP.

Operative methods

Operative methods of six patients were outlined in Table 2. The diagnosis of Amyand's hernia of six patients was obtained during surgery. Two patients whose laboratory examination was normal had normal appendixes, and inflamed appendixes were detected in the remaining four patients (Fig. 2). Case 3, and 6 were classified as type I Amyand's hernia whereas Case 1, 2, 4, and 5 were type II Amyand's hernia. Among four patients with type II Amyand's hernia, the appendix of Case 1, 2, and 5 was perforated and that of Case 4 was only inflamed. Two patients (Case 3 and 6) who had normal appendixes received a tension-free mesh repair through the inguinal incision. However, those with inflamed or perforated appendix received different surgical procedures. Case 1 underwent appendicectomy and the Bassini suture repair through the inguinal incision; Case 2 received appendicectomy and a tension-free mesh repair through the inguinal incision; Case 4 underwent laparoscopic appendicectomy and tension-free mesh repair; Case 5 received laparoscopic appendicectomy without further management of the hernia concerning the serious localized infection and inflammation. The drainage catheter was placed in the inguinal canal of most patients except Case 3.

Table 2
Operative methods

Case No.	Amyand's Hernia type	Appendix	Surgical approach	Appendicectomy	Herniorrhaphy technique	Drainage
1	II	Perforated	Inguinal incision	Yes	Bassini suture repair	Yes
2	II	Perforated	Inguinal incision	Yes	Tension-free mesh repair	Yes
3	I	Normal	Inguinal incision	No	Tension-free mesh repair	No
4	II	Inflamed	Laparoscopy	Yes	Tension-free mesh repair	Yes
5	II	Perforated	Laparoscopy	Yes	No repair	Yes
6	I	Normal	Inguinal incision	No	Tension-free mesh repair	Yes

Postoperative outcomes

Postoperative management and outcomes were detailed in Table 3. The patients received different antibiotics after surgery. Case 6 suffered from incision infection after surgery whereas other patients did not. No other postoperative complications were detected in six patients. The discharge time after surgery of six patients was 9.8 ± 6.1 days (range: 3–20 days). Case 3 who had normal appendix had the shortest hospitalization time after surgery, whereas Case 6 who had normal appendix but developed incision infection had the longest hospitalization time after surgery. Patients who received inguinal incision had a

longer median hospitalization time compared with those who received laparoscopy (10.8 ± 7.0 vs 8.0 ± 3.0 days). No patient suffered hernia recurrence during the follow-up.

Table 3
Postoperative outcomes

Case No.	Antibiotics after surgery	Surgical wound infection	Discharge time after surgery (days)	Deaths
1	Piperacillin sodium and sulbactam sodium combined with metronidazole	No	15	No
2	Piperacillin sodium and tazobactam sodium	No	5	No
3	Cefazolin sodium pentahydrate	No	3	No
4	Cefoperzone sodium and tazobactam sodium	No	5	No
5	Cefoperzone sodium and tazobactam sodium	No	11	No
6	Ceftriaxone combined with moxifloxacin	Yes	20	No

Discussion

The pathogenesis of Amyand's hernia associated acute appendicitis remains controversial. Previous studies indicated that muscle contractions or other sudden increases in intra-abdominal pressure might compress the appendix, resulting in inflammation [11, 12]. Moreover, an extraluminal obstruction of the appendix usually causes appendicitis due to pressure in the hernia neck rather than intraluminal obstruction [10, 11]. In our study, we presented six cases with Amyand's hernia, in which two had normal appendix and four had inflamed appendix. The different operative methods and outcomes of six patients would provide a reference for the treatment of Amyand's hernia.

A definitive preoperative diagnosis of Amyand's hernia is rare since the diagnosis is usually made during surgery. Physical examinations, laboratory examinations, and imaging examinations are not always helpful for the differential diagnosis of Amyand's hernia. With respect to imaging examinations, CT scanning can facilitate the diagnosis of Amyand's hernia. However, CT is usually not the first choice for an uncomplicated inguinal hernia [13]. Therefore, the diagnosis of Amyand's hernia will be missed at that time. Sonography has been reported as a valuable examination in the preoperative screening of Amyand's hernia since it is cheap and convenient for painful patients [14, 15]. Moreover, the suspected lesion can be further validated by CT. However, a preoperative diagnosis of Amyand's hernia based on ultrasound alone depends on the proficiency of the operator and, for that reason, remains a relatively unreliable imaging modality to accurately diagnose Amyand's hernia [16]. Therefore, laparoscopic surgery can function as a diagnostic and therapeutic approach. Recently, a systematic review indicated that CT

was the definitive diagnostic modality in patients with preoperative diagnosis.[17] In our six cases, four patients were diagnosed with appendicitis and inguinal hernia by CT whereas two patients were diagnosed with inguinal hernia by ultrasonography. However, the diagnosis of Amyand's hernia of these patients was obtained during surgery. In our opinion, CT imaging can facilitate the diagnosis of inguinal hernia but it is difficult to diagnose Amyand's hernia. As shown in Fig. 1, the density of the appendix is similar to that of the intestine and the identification of the appendix within the hernia is made after surgery. Therefore, CT is limited for the definite diagnosis of Amyand's hernia. The diagnosis of Amyand's hernia should be made by laparoscopy or laparotomy.

Losanoff and Basson have described four subtypes of Amyand's hernia and recommend different treatments (Table 4) [8]. Amyand's hernia with a normal appendix is classified as type I whereas type II-IV includes acute appendicitis. Therefore, Case 3 and 6 in our study were classified as type I Amyand's hernia and other cases were defined as type II. Generally, the primary management for Amyand's hernia with a non-inflamed appendix is hernia repair without appendectomy [7, 18, 19, 20]. Some clinicians believe that this will decrease the occurrence of postoperative complications because appendectomy will convert a clean surgery into a clean-contaminated surgery. Also, the remaining appendix can be further used to replace the extrahepatic biliary tract, perform urinary diversion, or conduct Malone procedure [21, 22]. Furthermore, during appendectomy, surgical manipulations in the base of the caecum may lead to the recurrence of inguinal hernias caused by the detachment of the deep inguinal ring [23]. Moreover, surgical manipulations involving the appendix might trigger secondary acute inflammation [22, 23]. However, these potential complications are minimized when the operation is performed laparoscopically [21, 22]. Shaknovsky et al. reported the successful treatment of an adult patient with type I Amyand's hernia after applying the Robotic platform Da Vinci Surgical System with 3D high-definition imaging [24]. Of the six cases included in this study, the treatment of Case 3 and 6 was tension-free mesh repair without appendectomy, which was consistent with the recommendation. As for other cases with type II Amyand's hernia, Case 1 and 5 received appendectomy without tension-free mesh repair, which was consistent with the recommendation. However, for Case 2 and 4 who had inflamed appendixes, the surgeons performed appendectomy and tension-free mesh repair and patients did not develop postoperative infections. This may be due to postoperative antibiotics and pelvic drainage.

Table 4
Four types of Amyand's Hernia

Classification	Description	Surgical management
Type 1	Normal appendix with an inguinal hernia	Hernia reduction, mesh repair, appendectomy in young patients
Type 2	Acute appendicitis within an inguinal hernia, no abdominal sepsis	Appendectomy through hernia, primary endogenous repair of hernia, no mesh
Type 3	Acute appendicitis within an inguinal hernia, abdominal wall, or peritoneal sepsis	Laparotomy, appendectomy, primary repair of hernia, no mesh
Type 4	Acute appendicitis within an inguinal hernia, related or unrelated abdominal pathology	Manage as types 1 to 3 hernia, investigate or treat second pathology as appropriate

Prosthetic mesh is typically contraindicated in patients with an inflamed or perforated appendix because of the increased risk for wound and mesh infections [9]. Besides, a recent study suggested that mesh repair should be conducted after removal of the appendix regarding an inflamed appendix without perforation or abscess. As for the perforated appendix, the synthetic mesh repair should be avoided. Moreover, mesh repair should be deferred if the inguinal canal had severe inflammation [17]. In our view, mesh repair is recommended when a non-inflamed appendix is discovered during herniorrhaphy. When acute appendicitis exists in the hernia sac, the surgeon should perform the appendectomy and tension-free hernia repair. In our study, the appendix of Case 4 is inflamed but not perforated, therefore, the mesh repair is applicable in this case. Case 1, 2, and 5 have perforated appendix, in which Case 1 and 5 do not receive mesh repair whereas Case 2 receives mesh repair. Since Case 2 did not develop postoperative infections, the application of the drainage tube and antibiotics may be helpful for the prevention of infections. Besides, the surgeon did not perform hernia repair in Case 5 because of the serious infection of the inner ring. In this case, it is better to perform two-stage surgery or one-stage reopen hernia repair. However, additional studies are required to determine the optimal surgical approaches for these patients.

Conclusions

Amyand's hernia is a rare presentation of inguinal hernias and the preoperative diagnosis of Amyand's hernia remains a challenge. CT and ultrasonography are helpful for the diagnosis but the definite diagnosis should be made by laparoscopy. The treatment of Amyand's hernia should be tailored based on the patient's condition and the type of Amyand's hernia. The application of tension-free mesh hernioplasty should be performed when the appendix is normally presented. If acute appendicitis exists in the hernia sac, an appendectomy should be performed and the application of prosthetic mesh should be avoided.

Declarations

Ethics approval and consent to participate

All methods were carried out in accordance with relevant guidelines and regulations. The study protocol, conform to the ethical guidelines of the 1975 Declaration of Helsinki, was approved by the Institutional Ethical Review Committee of The Dalian Municipal Central Hospital Of Dalian Medical University. Ethical approval was waived by the local Ethics Committee of The Dalian Municipal Central Hospital Of Dalian Medical University in view of the retrospective nature of the study and all the procedures being performed were part of the routine care. Written informed consents were obtained from all the participants.

Consent for publication

Informed consent was obtained from all individual participants included in the study.

The authors affirm that human research participants provided informed consent for publication of the images in Figures 1a,1b,1c,2a and 2b.The participant has consented to the submission of the case report to the journal.

Written informed consent for publication of their clinical details and/or clinical images was obtained from the patient/parent/guardian/ relative of the patient. A copy of the consent form is available for review by the Editor of this journal

Availability of data and materials

All data generated or analysed during this study are included in this published article.

Competing interests

There are no conflicts of interest to declare.

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Authors' contributions

Y.Gao and T.Zhang contributed equally to this work.Y.Gao—study conception and design, acquisition of data, analysis and interpretation of data, drafting of manuscript.T.Zhang—study conception and design,analysis and interpretation of data, critical revision of manuscript. M.Zhang—acquisition of data.Z.Hu—acquisition of data.Q.Li—acquisition of data. X.Zhang—study conception and design, critical revision of manuscript. All authors read and approved the final manuscript.

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Figures

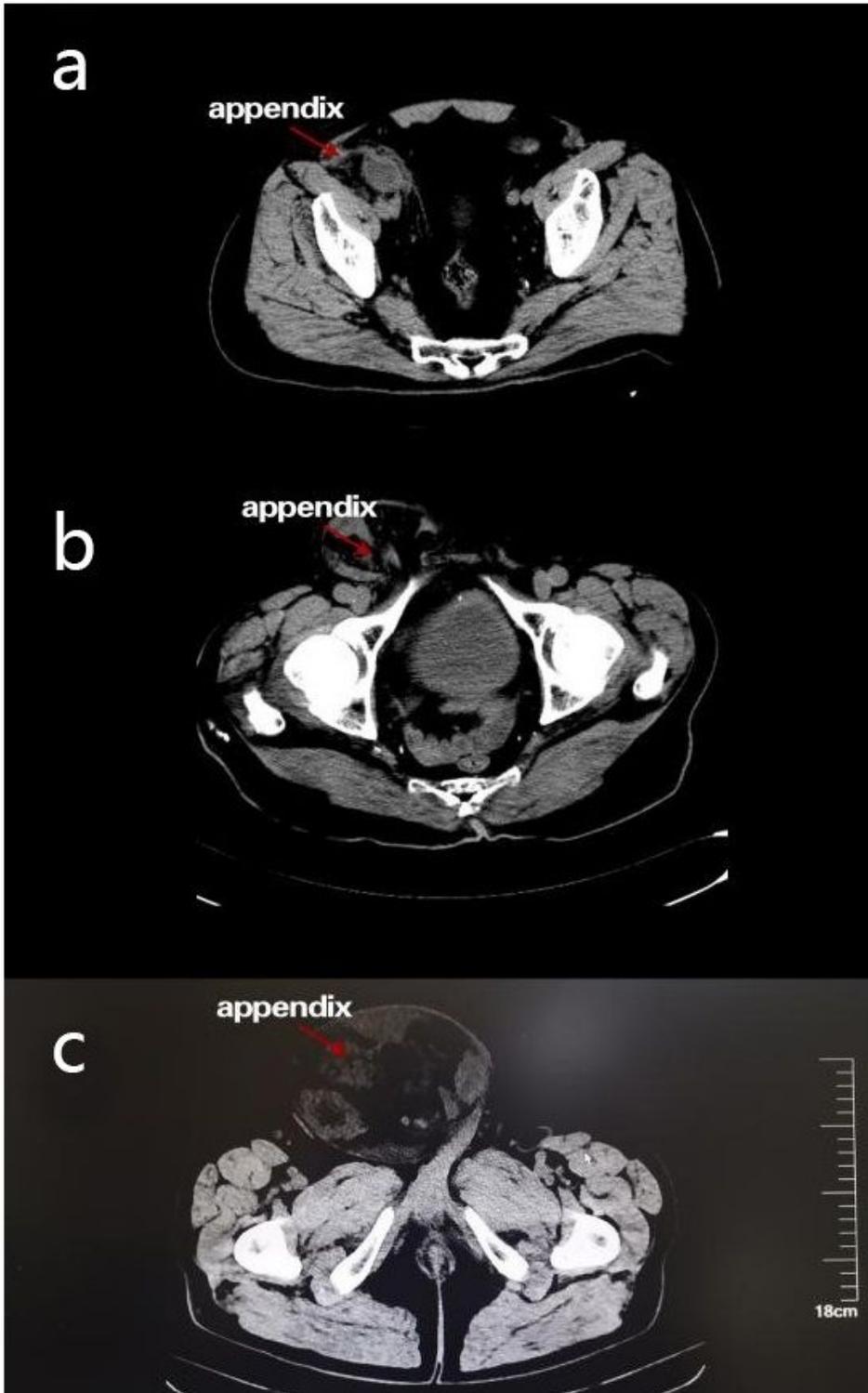


Figure 1

CT images of patients with Amyand's hernia. (a) Preoperative CT images of an inflamed appendix in Case 1. (b) Preoperative CT images indicated that the appendix was located in the hernia sac in Case 5. (c) Preoperative CT images indicated that the appendix was located in the hernia sac in Case 6.

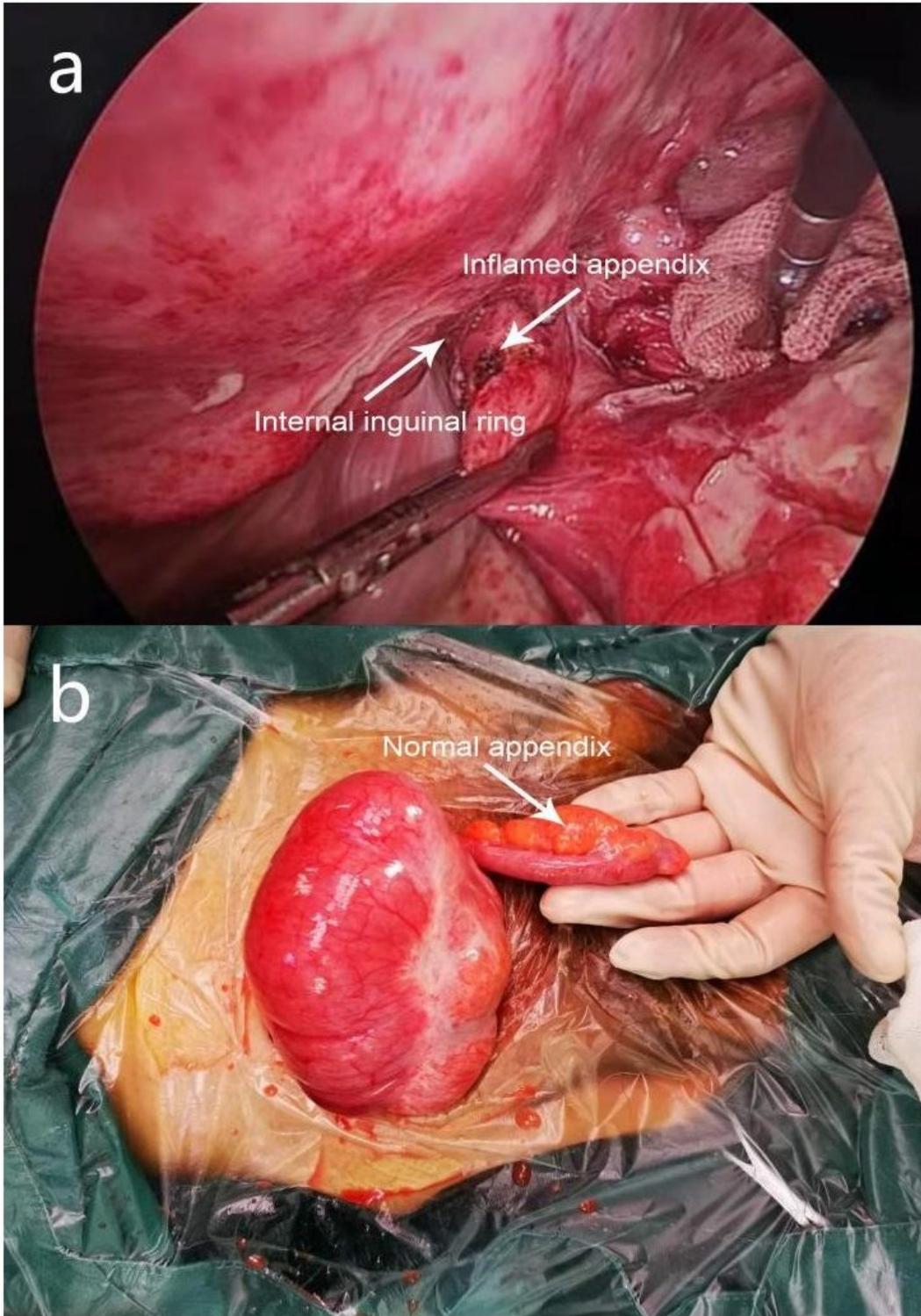


Figure 2

Appearance of normal or inflamed appendix discovered during surgery. (a) The inflamed appendix was separated from the hernia sac under the laparoscope in Case 5. (b) The cecum, appendix, and the distal portion of the ileum were contained in the hernia sac in Case 6.