Amyand´s hernia: a report of two cases and review of the bibliography

ABSTRACT

**Background:** Presence of the vermiform appendix in an inguinal hernia sac is an uncommon finding (1%) and is exceptionally rare if it is inflamed (0.13%). Clinically simulating incarcerated inguinal hernia and appropriate preoperative diagnosis is exceptional. We present two unusual cases of Amyand's hernia and review of the literature.

**Clinical cases:** Case 1. We present the case of a 78-year-old male with incarcerated right inguinal hernia. Diagnosis of Amyand's hernia was made preoperatively by abdominal computed tomography. Case 2. We present the case of an 82-year-old female with symptoms of an incarcerated right femoral hernia that finally showed an Amyand's hernia through a right inguinal hernia.

**Conclusions:** Amyand's hernia is a rare entity. Preoperative diagnosis is uncommon and should always be considered in the differential diagnosis in cases with clinical signs of incarcerated right inguinal hernia.

**Key words:** Amyand's hernia, acute appendicitis, incarcerated hernia.
BACKGROUND

An inguinal hernial sac can sometimes contain the urinary bladder, ovary, fallopian tube, colon with diverticulitis, or, as in the two cases referred because of unusual presentation of the vermiform appendix, in which case it is known as Amyand’s hernia. According to Hutchinson and Kueper et al., although DeGarangeot described it for the first time in 1731 the particular placement of a not inflamed cecal appendix in an indirect inguinal hernia, it was not until 1735 when Claudius Amyand carried out the first appendectomy. This surgeon, co-founder of the St. George’s Hospital and forerunner of the vaccination against smallpox, operated on an 11-year-old child who suffered from an inguinoscrotal hernia complicated with a stercoral fistula. During the surgical intervention the perforated appendix was seen within the hernia sac and the first appendectomy was successfully performed, as described in the scientific literature, through an inguinal access.1,2

The existence of a vermiform appendix in an inguinal hernia sac is an uncommon finding (1%), very rare if it is found to be inflamed (0.13%) and almost always described in males. From the clinical point of view it simulates an incarcerated hernia. Very few times is the diagnosis adequately established before the operation.3,4

Two unusual cases of Amyand hernias are reported and the literature is reviewed.

CLINICAL CASES

Case 1

A 78-year-old male, with history of type 2 diabetes mellitus on treatment with oral antidiabetics and chronic bronchitis presented to the Emergency Department due to generalized abdominal pain, nausea and vomiting of 24-h evolution. On physical examination an indurated tumor was appreciated in the right inguinal region, which was painful to palpation and not reducible, compatible with incarcerated inguinal hernia. The abdomen was found to be distended, and deep palpation of the hypogastrium elicited signs of peritoneal irritation. Laboratory results were within normal limits: 7100 leukocytes/mm³, neutrophils 77.5%, hemoglobin 12.6 g/dl, hematocrit 38.2% and 266,000 platelets/mm³. On computed tomography (CT) of the abdomen, acute appendicitis was noted with incarcerated hernia contents, without intraabdominal free fluid (Figures 1 and 2).

Clinical findings and complementary tests led to the diagnosis of Amyand’s hernia; therefore, the patient was urgently surgically intervened. The patient received antibiotic prophylaxis with gentamicin and metronidazole. The approach was inguinal and the suspected diagnosis was confirmed with the finding of a gangrenous appendicitis with inflammatory exudate as contents of the hernia sac in the indirect inguinal hernia. Due to the advanced inflammatory process and high risk of infection it was decided to repair the hernia with a herniorrhaphy with the Bassini technique with Prolene sutures (Surgilene®, Covidien AG). The

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Figure 1. Frontal view of the preoperative abdominal computed tomography (CT) of the male patient with an Amyand hernia. The appendix is inflamed in the right inguinal canal (white arrow).
patient progressed favorably and was discharged on the 6th postoperative day. Histopathological report showed an acute gangrenous appendicitis. Currently, after 2 years of follow-up the patient is asymptomatic with signs of hernia recurrence.

Case 2

An 82-year-old female patient is presented with personal history of high blood pressure and atrial fibrillation being treated with doxazosyn, acenocumaron and digoxin. She was seen in the Emergency Department because of pubic pain of 3 months duration, which increased during the last 24 h. She was afebrile and did not report abdominal pain. On physical examination, an erythematous tumor was noted in the right pubic region with plaques of necrosis and extensive edema up to the right labia majora. It was very painful to palpation, suggestive of a complicated right crural hernia. Plain x-ray of the abdomen did not demonstrate any pathological findings. Laboratory studies reported leukocytosis with 13,100 leukocytes/mm³, neutrophils 78.5%, hemoglobin 12.4 g/dl, hematocrit 37.7%, and platelets 250,000/mm³. Prothrombin activity was 8.9%, which was corrected with the administration of Factor IX as complex II-VII-IX-X. It was decided to operate on the patient urgently via a right inguinal approach. During the surgery an abscess limited to the subcutaneous cellular tissue was found, secondary to perforation of the hernial sac due to a perforated gangrenous appendicitis at the tip and contained in the sac that finally formed part of a direct inguinal hernia. Appendectomy and right inguinal herniorrhaphy was carried out according to the Bassini technique. Because of the adjacent purulent exudate, Prolene suture was used (Sur-gilene© Covidien AG). Diagnosis of Amyand’s hernia was established and required antibiotic treatment and wound care until resolution of the surgical wound infection. The pathological study confirmed the diagnosis of perforated acute gangrenous appendicitis at the tip. After a year of follow up, the patient has had no hernia recurrence.

DISCUSSION

Amyand hernia, defined by Fernando and Leelaratna as an inguinal hernia whose sac may contain a) normal cecal appendix, b) inflamed appendix, or c) perforated appendix, constitutes an uncommon problem. Losanoff and Basson classified it into four types: type 1: normal appendix; type 2: acute appendicitis; type 3: acute appendicitis and peritonitis and type 4: acute appendicitis and another abdominal infection.

It is more common in males and a wide age range has been described (from 3 weeks up to 89 years). Amyand hernia almost always occurs on the right side because of the anatomic position of the cecal appendix; however, in some cases of situs inversus, intestinal malrotation and mobile cecum, it can present itself on the left side. Our second case is unusual due to the finding of a...
gangrenous appendicitis in an inguinal hernia in a female.

The etiological theories place this disorder as a finding, or on the contrary, as a relationship between the appendicular incarceration and the inflammation. The majority of authors defend the second hypothesis, which suggests that the entrance of the cecal appendix in the hernia sac exposes it to trauma that would lead to intrasacular adhesions. Continued trauma and muscular contractions of the abdominal wall would reduce or suppress the appendicular blood flow, which favors an acute appendicitis.8,10,11

The clinical manifestations described in the literature report a typical picture of appendicitis with epigastric or periumbilical pain radiating to the right iliac fossa. The pain is characteristically colicky, compared with the dull and continuous pain of a strangulated intestine. Fever and leukocytosis are not constant. On the face of an incarcerated right inguinal hernia these manifestations make adequate preoperative diagnosis difficult.3,9

Preoperative abdominal echography and CT are the most useful radiological tests for diagnosis; however, they are not routinely performed in most cases.3,8,11 Only a high grade of suspicion in a patient with an incarcerated right inguinal hernia with signs of local peritonitis and without clinical signs of intestinal obstruction would lead us to performing an echography or abdominal CT that allow establishing the correct preoperative diagnosis. Our first case stands out as exceptional with a preoperative diagnosis by abdominal CT, which has been described in a few cases in the literature.12-14

Differential diagnosis should be established with a strangulated hernia, strangulated omental appendix, Ritcher hernia, testicular torsion, orchioepididymitis, hemorrhagic testicular tumor, acute hydrocele and inguinal lymphadenitis.3,8 The surgical procedure of choice is appendectomy and hernia repair when it is found to be inflamed. There is controversy in relation with respect to the appropriate time and technical intervention of the hernia. According to the Losanoff and Basson classification,6 what is recommended for type 1 hernias is appendectomy and hernioplasty with access at the inguinal level or even laparoscopically.15,16 For type 2 appendectomies the inguinal route with herniorrhaphy and for types 3 and 4 appendectomy with laparoscopic abdominal exploration and inguinal herniorrhaphy are recommended. For the last three types, herniorrhaphy is the most recommended technique for hernia repair due to the high risk of infectious complications by the abdominal septic process implied by performing a hernioplasty.3,8,9

With adequate perioperative treatment, mortality due to Amyand’s hernia has been reduced from 14-30% to 5.5% in recent series. Surgical wound infection is the most common complication followed by hernia recurrence after herniorrhaphy (4.9-7.6%), intra-abdominal abscess and necrotizing fasciitis if surgical treatment is delayed.3,7,9

In conclusion, Amyand’s hernia is an exceptional condition with a rare preoperative diagnosis, which must always be considered in the differential diagnosis in patients with clinical signs of incarcerated right inguinal hernia.

REFERENCES