Abstract

Introduction: The presence of vermiform appendix inside the hernia sac is infrequent. In the literature, the reported incidence is of about 1% of all hernias. The finding of acute appendicitis in the inguinal hernia is even rarer, clinical picture that is called Amyand’s hernia. Another possible situation is the Richter’s hernia, where part of the circumference of the intestine is trapped and strangled in the hernia hole, leading to ischemia, gangrene and visceral perforation. We report a case presented at the Hospital Regional do Cariri in Juazeiro do Norte-Ceará, of Amyand’s hernia associated with Richter’s hernia in the same hernia sac, nosological entity of rare occurrence, followed by review of the available literature about the topic.

Method: The research was conducted on PubMed using the un-terms “Amyand’s hernia” and “Richter’s hernia” then the most relevant publications about the topic were filtered and correlated with the findings of the case reported.

Discussion: In this type of inguinal hernia, the hernia content is constituted by the vermiform appendix concomitant to the antimesenteric pinched edge of the ileal loop. There was still the incidental finding of a Meckel’s diverticulum. The patient had a clinical diagnosis of incarcerated hernia and was submitted to surgical treatment with the finding of Amyand’s hernia associated with Richter’s hernia, evolving satisfactorily in the postoperative.

Keywords
Amyand’s hernia, Richter’s hernia, appendectomy, Meckel’s diverticulum.

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Introduction
The presence of the vermiform appendix inside the hernia sac is infrequent. In the literature, the reported incidence is of around 1%. The finding of acute appendicitis in the inguinal hernia is even rarer. When we find the vermiform appendix as a content of the inguinal hernia sac, inflamed or not, the clinical picture is called Amyand’s hernia. This type of hernia is more common in men, and the preoperative diagnosis is difficult [1].

Richter’s hernia is a type of hernia in which only part of the circumference of the intestine is trapped and strangulated in the hernial orifice leading to ischemia, gangrene and visceral perforation. The portion of the intestine that is most commonly involved is the distal ileum; however, any portion of the gastrointestinal tract can become incarcerated. Richter’s hernia occurs when the size of the hernial orifice is large enough to retain part of the circumference of the intestinal wall, but prevents the exit of the intestinal loop. The Richter’s hernia progresses more rapidly to gangrene due to the compromised blood supply [2].

Meckel’s diverticulum is one of the most common congenital abnormalities of the gastrointestinal tract and it occurs in approximately 2% of the population. This true diverticulum results from an incomplete obliteration of the vitelline duct during the fifth week of gestation and emerges from the antimesenteric border of the distal ileum. In most cases, it is asymptomatic and the diagnosis occurs occasionally during a surgery performed because of other intra-abdominal disorders [3].

The treatment of the inguinal hernia, even the non-incarcerated, is eminently surgical (safeguarding pathological conditions of prohibitive base to the procedure) becoming of emergency in the case of its strangulation. The presence of the appendix and/or antimesenteric border of the small intestine in the hernia contents may change its evolution; it may become drastic [4].

When faced with a case of Amyand’s hernia associated with Richter’s hernia, we feel the need to add to the literature another occurrence of this nosological entity and to discuss the pathophysiology, diagnosis and treatment of this rare type of hernia [5].

Method
An extensive bibliographic research was conducted in worldwide literature covering scattered case reports, literature reviews and original articles published from 1928 to 2013, written in English, Portuguese and Spanish. [6]

The research was conducted between the months of January and February 2015, with literature acquired through electronic means, in the databases of these sites: Biblioteca Virtual em Saúde (BVS), Scientific Electronic Library Online (SciELO), PubMed and Medline. The descriptors used were: hérnia de Amyand, Amyand’s hernia.

Results
47 case reports were found in the search performed in databases using the keywords: “hernia” and “Amyand”. In the search for “hernia” and “Richter” 121 articles were found. When the following terms were used: “hernia”, “Amyand” and “Richter”, no cases were found. Articles were analyzed for discussion.

Case Report
The patient, male, was admitted for the first time in the emergency sector of the Hospital Regional do Cariri in 15/07/2014, at approximately 11:45 AM. At the time of the treatment, he was 69 years old. He arrived complaining of severe abdominal pain, with a sudden onset, early in the morning, not knowing the precise time, being initially assessed by a general practitioner and then an evaluation was requested from the general surgeon. At the admission, physical examination was performed and pain to deep
Palpation was found especially in the right iliac fossa and periumbilical region, without signs of peritoneal irritation. At the examination, right inguinal hernia irreducible to the maneuvers was also noted, even after analgesia, but very painful. On the herniation site, exuberant and phlogistic signs were not noticed. Patient denied incarceration until the night that preceded the surgery, being then sent to the surgical center.

Patient was submitted to surgery, which started by right inguinotomy, under spinal anesthesia. After dissection of the plans, the hernial sac derived from mixed inguinal hernia was evidenced (Nyhus classification 3b), with blackish content of probable necrotic nature, which later revealed itself as a vermiform appendix apex (distal 1/3) necrotic and also clamping of almost all circumference (> 2/3) of the small intestine loop and of blackish color too (Figure 1).

Faced with this finding, a conversion of the anesthetic and conduction of an exploratory laparotomy by trans-umbilical midline incision was chosen, for better evaluation and field exposure and also for a safer correction and anastomosis.

After opening the abdominal cavity, the hernia content was reduced and the following was evidenced: distal third necrosis (about 5 cm of its full 15 cm) of appendix and necrosis in a pyramidal shape (being the apex of the pyramid, the mesenteric border) of the ileal loop, lying approximately 2 meters and fifty centimeters from the ileocecal valve (Figure 2).

During inventory of the abdominal cavity, there was evidence of Meckel's diverticulum, of base with diameter of approximately 2 cm, lying approximately 1 meter away from the ileocecal valve (Figure 3).

Faced with this clinical picture, there was the decision to perform an appendectomy with subsequent burial of appendicular stump in a tobacco pouch made with prolene 3-0. There was also the performance of an enterectomy of approximately 15 cm, being located at its center the necrotic area derived...
from hernia content, with reconstruction by entero-entero anastomosis end to end made in a single plane with continuous suture with prolene 3-0. A diverticulectomy of Meckel’s diverticulum was also realized, with wedged section of its base and cross enterorrhaphy in single layer with continuous suture with prolene 3-0.

An anterior abdominal wall closure with nylon 0 double and subsequent correction of right inguinal hernia defect by the Bassini technique was performed. We opted for the non-use of polypropylene mesh because of the local gross contamination caused by both the appendix and by the necrotic ileal loop.

**Antibiotic therapy was used with ciprofloxacin and metronidazole**

Patient evolved in the postoperative with paralytic ileus, responding appropriately to the electrolyte support, symptomatic (especially analgesics and antiemetics), tolerating well the progressive increase in the diet. An accumulation of serous fluid in surgical wound was also observed, treated by opening stitches in skin to drain collection and subsequent healing by secondary intention aided with the use of Fibrase® ointment.

Discharge was given on the eighth postoperative day and ambulatory discharge on the fortieth day, with surgical wound at an advanced stage of healing, being referred to a primary care unit for follow-up until fully healed. In the period after hospital discharge, he sought the emergency sector of the hospital once because of vomiting episodes, being successfully treated only with symptomatics.

The results of the histopathological examination showed no signs of malignancy in the appendix or hernial sac, confirmed hemorrhagic infarction of all layers of the small intestine and the nature of true diverticulum of Meckel’s diverticulum.

**Discussion**

A curious fact is the rare possibility of appendiceal incarceration being located in a left inguinal hernia, in cases of long appendices, cecum with increased mobility, poor intestinal rotation or situs inversus [7].

There is a very disparate variety of epidemiological data found in the literature with regards to Amyand’s hernia: the incidence varies from 0.19% to 1.71% of the cases of inguinal hernias considering the normal appendix, and even rarer, 0.07 to 0.13%, if we consider the incidence of acute appendicitis inside the inguinal hernia sac. The incidence rate in infants is 3 times higher than in adults. There are few cases of the Amyand’s hernia in women, and in this population it tends to occur in postmenopausal period. The mortality rate ranges from 5.5% to 30% and is mainly due to the complications, especially abdominal sepsis. [8]

The preoperative diagnosis of the Amyand’s hernia is not simple, and it is usually an incidental finding during surgery [9]. Physical examination, laboratory exams and imaging tests are not always useful in the differential diagnosis. The most common complaints include sudden epigastric or periumbilical pain with increased sensitivity located in the right lower quadrant of the abdomen, combined with irreducible mass in the inguinal or inguinal-scrotal area. [10] This presentation however, often gives the clinical impression of a strangulated hernia, making the clinical diagnosis of Amyand’s hernia difficult. [11]

In making the comparison of the clinical case with the consulted literature, we came across several parallels and a few facts fleeing from the general rule. Firstly, the clinical presentation was very similar to that described in most cases: irreducible hernia with abrupt onset of pain by weight in periumbilical region and right lower quadrant. The already advanced age, flees from the infant standard of the majority. [12]
Another fact parallel to most literature cases was that our diagnosis was given in the intraoperative, because the clinical history and physical examination were sufficient for the indication of surgery. We consider that additional tests would only be costly to the hospital and risky to the patient because they would postpone the surgical treatment, increasing morbidity and the risk of complications: the infection, at that time restricted to the sac and hernia defect, could enter the abdominal cavity [13].

An interesting fact caught our attention: even though the patient has Richter’s hernia in the same hernia defect, with more than 2/3 of its light obstructed, there were no complaints compatible with intestinal obstruction or abdominal distension in the preoperative physical examination or upstream distention of the bowel loops in the intraoperative findings. This fact is considered in favor of incarceration (and subsequent strangulation), at least of the small intestine loop, being acute, corroborating with a story of approximately 6 hours counted by the patient. [14]

The surgery was performed as directed by the largest portion of the authors. After inguinotomy and verification of necrosis and the suspicion on our part of continuity of the infection process into the abdominal cavity and the very narrow field to perform appendectomy and enterectomy because of the hernia defect, we proceeded with laparotomy by midline incision. The hernia defect was corrected by using the Bassini technique instead of Shouldice technique, as oriented by the majority of the authors, because of the greater experience of the surgeons involved with this technique, according to the literature, the hernia recurrence rate is inversely proportional to the degree of familiarity of the surgeon with the technique used. [15-17]

Conclusion
As in most series, there was no infection of the surgical wound. As a complication, there was only paralytic ileus, which postponed the discharge of the patient to eight days.

Ethical aspects
The ethical aspects were respected maintaining the confidentiality of the information contained in the medical records, which were used exclusively for the scientific publication, preserving the anonymity of the patient's identity.

The patient authorized the publication of the case and signed the Informed Consent.

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