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CASE REPORT

LEFT AMYAND'S HERNIA IN A 1-YEAR-OLD MALE INFANT: REPORT OF A RARE CASE IN COMMON PRESENTATION

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Abstract. *Amyand's hernia is a rare type of hernia with the appendix and cecum trapped in a hernial sac. A one-year-old male infant was referred with irreducible left inguinal hernia for about 1 week. No signs of intestinal obstruction were presented, and pain was tolerated. A herniotomy was performed on the patient. Intraoperatively, the appendix and cecum were found in the left hernial sac, which this rare presentation named left Amyand's hernia. Cecum and appendix were considered normal. No perforation and significant inflammation were found; hence, no appendectomy was performed. Left Amyand's hernia is a very rare case, mostly in pediatric surgery, in which the current decision of diagnosis-making can only be made intraoperatively.*

Key words: *amyand's hernia, appendix, herniotomy, inguinal hernia*

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INTRODUCTION

Amyand's hernia is a rare type of inguinal hernia named after a French surgeon, Claudius Amyand, who first described a case of hernia with the cecum and appendix entrapped in it and conducted an appendectomy through herniotomy [1]. Reports regarding this case are currently limited, but incidence is estimated at 0.5-1% cases of inguinal hernia and 0.07-0.13% cases of appendicitis [2]. Amyand's hernia cases are mostly reported in bimodally, in groups of infancy and elderly, respectively. Diagnosis of Amyand's hernia is mostly reported as an accidental discovery during surgery. Some cases were reported with appendectomy conducted from the hernial sac incision, while some others preferred to conserve the appendix [3]. This rare case of Amyand's hernia occurred in Indonesia, which presented as a common presentation of inguinal hernia found in an infant.

CASE REPORT

A 1-year-old male infant was referred to a public hospital with an enlarged mass at the left inguinal region and left hemiscrotum and has been irreducible since the previous week. The size of the mass was approximately 6 x 3.5 cm, with a scrotum circumference

of 7.6 cm. Parents reported that the mass reduced spontaneously when the patient fell asleep and bulged whenever the patient cried. No other medical or surgical history was reported.

Physical examination revealed no abdominal distension, no nausea and vomiting, no feeding problem, and normal peristaltic sound. There were no signs of obstruction or strangulation of the bowel presented, and the patient was initially diagnosed with an irreducible left inguinal hernia.

The patient was then prepared for an elective herniotomy. Preoperative laboratory examination and chest X-ray were considered normal. An inguinal ultrasound examination was conducted prior to surgery, as depicted in Figure 1. We identified a lumen-like structure with a narrowing area in the inguinal canal, which is suspected to be an appendix.

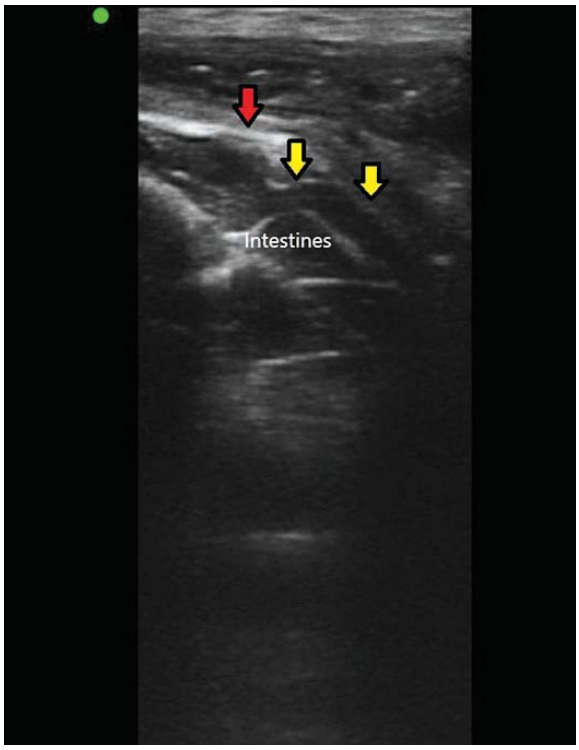

Fig. 1. Ultrasound examination of the left inguinal hernia, bowel (red arrow) and appendix (yellow arrow) observed

Surgery was conducted under general anesthesia. Upon surgery, the hernia remained irreducible. Hence, the incision at the external inguinal annulus was performed to slightly reduce the bowel in the hernial sac. As the patient processus vaginalis was dissected, a long appendix and distal part of the caecum were found in the hernial sac, as pictured in Figure 2. The structure of the appendix was mobile, smooth, and viable, and no signs of inflammation or perforation were presented (yellow arrow). Thus, this patient was diagnosed with type 1 left Amyand's hernia.

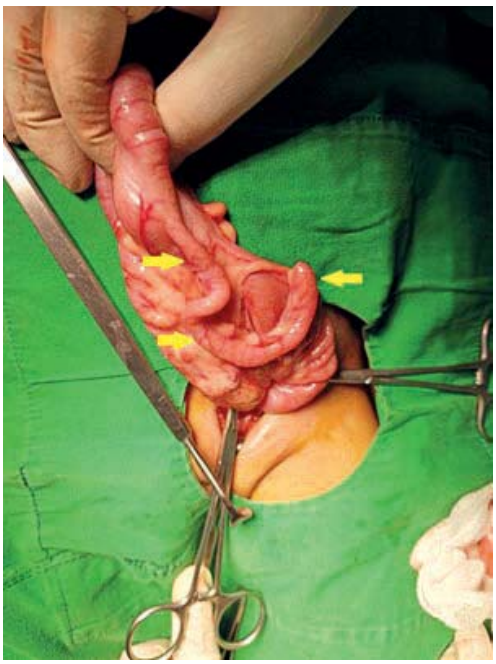

Fig. 2. Intraoperative appearance of the vermiform appendix (yellow arrow)

We considered that no significant inflammation or injury occurred to the appendix and cecum; hence, no appendectomy was performed. The cecum and appendix were reduced manually. The proximal patent processus vaginalis was then ligated at the level of preperitoneal fat with double ligation using absorbable polyglactin 3/0 suture, and the external annulus was repaired. The surgery procedure went without complication, and the post-operative care of the patient was considered safe. The patient received a broad-spectrum antibiotic with cefotaxime and paracetamol as analgetic. The patient was discharged 3 days after surgery and had no other complaints.

DISCUSSION

Amyand's hernia was reported in approximately only 1% of all cases of inguinal hernia and coincidentally found with appendicitis inside the hernial sac, which was estimated at 0.1% of all inguinal hernia cases. This type of hernia was recorded to occur more among children and the late elderly, which presented as a bimodal age distribution in epidemiologic studies. Children and the late elderly are at 3 times higher risk compared to those in a group of young adults [1, 2].

The pathophysiology and mechanism of Amyand's hernia remains unknown. Some reports hypothesized that the occurrence of Amyand's hernia is associated with the presence of a fibrous connection between the appendix and testis during testicular descent. This connection causes the appendix to ad-

here to the patent processus vaginalis as the testis enters the scrotum. This mechanism occurs at the early organ development of the appendix and testis, which both start in the abdominal cavity. This theory may be possible in cases of Amyand's hernia in children, which is mostly congenital, while cases in the elderly are related to abdominal wall weakness and loosening of mesenteric tissue, which anchors the appendix [2, 3].

Most of Amyand's hernia cases occurred in the right inguinal hernia, which is due to the anatomical structure of the appendix and cecum in the right lower quadrant of the abdomen. This position is very likely for the appendix and cecum to be connected and trapped in the right inguinal hernia. Left Amyand's hernia, as described in this case, is very rare, reported only 4% of all Amyand's hernia cases [4]. The presence of the appendix in the left inguinal can be stated as an anatomical anomaly that may occur in cases such as situs inversus, intestinal malrotation, and mobile cecum where the appendix is located abnormally. Situs inversus can be easily diagnosed with the whole anatomical structure presented inversed on the opposite side and can be suspected from a preoperative thorax radiograph, while malrotation may require further radiological examination with a contrast study, which was not performed in this case as there was no further treatment needed due to no associated symptoms presented. Mobile cecum with loose anchoring of cecum and appendix from the mesentery is the most likely to cause left Amyand's hernia when other anomalies are not observed [5, 6].

Amyand's hernia is usually diagnosed intraoperatively when opening the hernial sac in a herniotomy. Currently, several imaging modalities are available to predict the presence of appendix and cecum in the hernial sac. Ultrasonography (US) is a widely known modality and is commonly used in determining the content of hernial sac in most hernia cases. In this case, we observed a narrow lumen structure at a distal end of the bowel structure in the hernial sac. Consequently, Amyand's hernia founding may be predicted through the US. However, operator experience has always been a weakness in US examinations, so this might not be the most suitable and appropriate measure for diagnosing Amyand's hernia [7]. Computed Tomography (CT) may be superior to diagnose Amyand's hernia, moreover with the adjunction of intraluminal contrast. This may visualize the presence of the appendix in the hernia sac. The controversy of CT in the early detection of Amyand's hernia is related to the clinical significance of further treatment since patients with or without the appendix in the hernial sac are commonly mandated to undergo herniotomy. In addition, the diagnosis will be discovered as the structures are found intraoperatively, and there are no preoperative treatment differences in patients with Amyand's hernia and other types of inguinal hernia [7, 8].

Treatment and surgical procedures in Amyand's hernia depend on its classification. Losanoff and Basson previously classified Amyand's hernia in 2007 into 4 types, which may determine the surgical procedure to undergo after an intraoperative finding of the appendix in the hernial sac is made. Type 1 Amyand's hernia diagnosis is made when the appendix is considered normal clinically. Type 2 may be stated when signs of acute appendicitis present symptomatically or when clinical intraoperative appearance without any signs of perforation is found. The third type of Amyand's hernia occurs when acute appendicitis is present in an inguinal hernia followed by signs of perforation or abdominal wall symptoms or peritoneal sepsis, whilst lastly, the fourth type is the third type of Amyand's hernia followed with other related or unrelated abdominal pathology or accompanied by the presence of ileus [8, 9].

This patient was admitted with a chief complaint of an inguinal lump suggestive of an inguinal hernia. Signs and symptoms of appendicitis regarding abdominal pain, nausea and vomiting, fever, and McBurney signs were not presented at all and clinically can be considered as the type 1 hernia. However, intraoperative findings presented no signs of acute inflammation and perforation suggestive for the type 1 hernia.

Current recommendation for the intervention in this case of type 1 still remains controversial. In contrast, while type 2 Amyand's hernia is recommended to undergo an appendectomy through herniotomy, and both type 3 and 4 are recommended to undergo an appendectomy through laparotomy due to surgical emergency to prevent progression of infection and sepsis, the type 1 Amyand's hernia can be treated with or without appendectomy. Some clinicians prefer to conduct appendectomy through herniotomy incision due to the consideration of preventing further episodes of acute appendicitis, especially in infants and pediatric patients. On the other hand, other surgeons prefer to conserve the appendix as clinically considered normal (type 1) to prevent the risk of infection coming from the herniotomy incision. Different pieces of evidence to conserve the appendix are based on the risk of hernia recurrency and requirement of mesh, as the manipulation during appendectomy through herniotomy may lead to the pressure and enlargement of the internal inguinal ring [8, 9].

We reported a case of type 1 Amyand's hernia, as though the appendix was considered long. However, there were no signs of acute appendicitis. A previous similar case at a comparable age was reported by Supangat et al., but the presentation was different: with signs of left incarcerated hernia. Intraoperative procedure revealed a left Amyand's hernia with an inflamed appendix and cecum, so type 2 was diagnosed, and an appendectomy was performed [10]. Another case was reported by Joshi et al. with left irreducible inguinal hernia, which revealed a type 1 Amyand's hernia during surgery. The patient was managed by conserving the appendix reducing it back into the abdominal cavity. The patient only had a herniotomy. The report followed up the patient with barium meal follow-through to prove the presence of situs inversus or intestinal malrotation, yet a normal finding was proved. Similar to this case, but due to limited facility and patient consent, follow-up of barium meal follow-through was not performed. Identifying the cause of the left-sided Amyand's hernia is due to intestinal malformation with mobile caecum, and it is not urgent or mandatory as the presence of intestinal malrotation without pathological signs and symptoms would require no further intervention [6].

CONCLUSION

Amyand's hernia is an uncommon type of hernia with the appendix trapped inside the hernia sac, but it may be missed in the diagnosis as there are no clinical differences from other types of inguinal hernia. A left Amyand's hernia is even rarer and associated with some anatomical anomalies, which makes it possible for the appendix to appear in the left inguinal canal. This rare case was suspected with early imaging study with inguinal ultrasound prior to surgery, as the appendix may be visualized as a narrow intestinal luminal-like structure. We reported a case of Amyand's hernia with the normal appendix that was presented and successfully treated without appendectomy, as conservative treatment can be considered whenever no perforation is present in the appendix.

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