

# Left Amyand's hernia associated with omphalocele

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## ABSTRACT

**Introduction.** Amyand's hernia is defined as the presence of the cecal appendix inside the hernia sac. It is usually located on the right side, but left-sided cases due to intestinal malrotation have also been reported.

**Case presentation.** 3-month-old male infant diagnosed with omphalocele at the 12<sup>th</sup> week of gestation undergoing surgery 48 hours post-birth. Bilateral inguinal hernia was diagnosed during follow-up, so scheduled bilateral herniotomy was performed. During surgery, the cecal appendix was identified inside the left herniary sac, so prophylactic appendectomy was carried out.

**Conclusions.** The incidence of Amyand's hernia ranges from 0.4 to 1% – three times higher in pediatric patients. Left-sided hernias are often associated with intestinal rotation disorders. Treatment of right Amyand's hernia is described in Losanoff-Basson's classification. In left-sided hernias, prophylactic appendectomy is recommended to avoid diagnostic confusion in case of appendicitis.

**KEY WORDS:** Indirect inguinal hernia; Appendectomy; Omphalocele.

## HERNIA DE AMYAND IZQUIERDA ASOCIADA A ONFALOCELE

### RESUMEN

**Introducción.** La hernia de Amyand consiste en la presencia del apéndice cecal en el saco herniario, generalmente en localización derecha, aunque se recogen casos de localización izquierda debido a malrotación intestinal.

**Exposición del caso.** Lactante varón de 3 meses de edad, con omphalocele diagnosticado en la 12<sup>a</sup> semana de gestación e intervenido a las 48 horas del nacimiento. Durante el seguimiento se diagnostica de hernia inguinal bilateral, realizando herniotomía bilateral programada. En la intervención se encuentra el apéndice cecal en el saco herniario izquierdo, procediendo a una apendicectomía profiláctica.

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**Comentarios.** La incidencia de la hernia de Amyand es del 0,4-1% (3 veces más frecuente en población pediátrica). La localización izquierda suele asociarse a alteraciones en la rotación intestinal. El tratamiento de las hernias de Amyand derechas se recoge en la clasificación de Losanoff-Basson. En las izquierdas, se recomienda realizar apendicectomía profiláctica para evitar confusiones diagnósticas en caso de apendicitis.

**PALABRAS CLAVE:** Hernia inguinal indirecta; Apendicectomía; Onfalocele.

## INTRODUCTION

Amyand's hernia is defined as the presence of the cecal appendix inside an inguinal hernia, most frequently on the right side owing its anatomical location.

Amyand's hernia was named after Claudius Amyand. It was first described in 1735 when Claudius Amyand performed the first appendectomy in an inguinal hernia in an 11-year-old boy.

We report the case of a left-sided Amyand's hernia<sup>(1-3)</sup>.

## CLINICAL CASE

The patient was a 3-month-old male infant, diagnosed with omphalocele pre-natally, at the 12<sup>th</sup> week of gestation (Fig. 1). His 35-year-old mother had no significant pathological or gynecological history. She was nulliparous and she used to smoke 5 cigarettes a day.

48 hours post-birth, once other associated malformations had been ruled out, the 63 x 65 x 55 mm (30 cm<sup>3</sup>) abdominal wall defect –filled with hepatic content primarily– was repaired (direct closure). No significant complications arose postoperatively (delayed digestive tolerance initiation with conservative management and adequate intake from day 11 post-surgery), so the patient was discharged (Fig. 2).



**Figure 1.** Pre-birth ultrasound imaging at the 14<sup>th</sup> week of gestation.



**Figure 2.** Omphalocele 48 hours post-birth.

At consultation follow-up, the patient was diagnosed with large indirect bilateral inguinal hernia, so bilateral herniotomy and omphaloplasty were scheduled.

At surgery –performed through modified Ferguson technique –, the ileocecal segment and the appendix were identified inside the left hernia sac. The cecal appendix had a normal macroscopic appearance, with no inflammatory signs, so prophylactic appendectomy was carried out (Fig. 3).

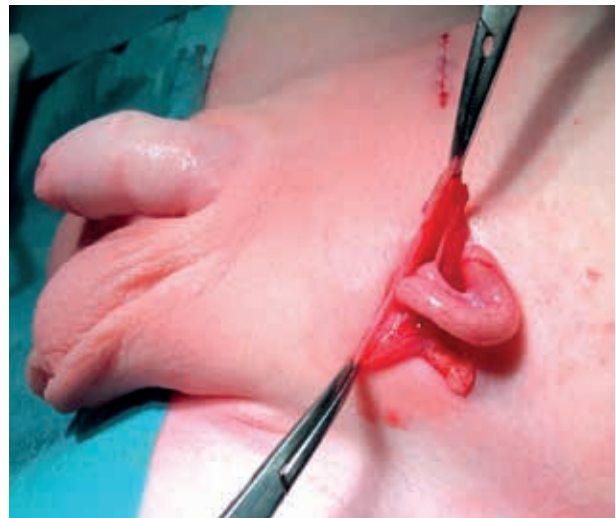
At postoperative follow-up, one month post-surgery, the injuries had a nice appearance in terms of healing, with the left spermatic cord still being slightly thickened.

## DISCUSSION

Along with De Garengeot's hernia (cecal appendix inside a crural hernia) and Littre's hernia (Meckel diverticulum inside a hernia sac), Amyand's hernia is one of the so-called rare hernias, which have a low incidence (0.4-1%). However, it can be up to three times more frequent in the pediatric population owing to persisting vaginal process<sup>(1,2)</sup>. Appendicitis associated with Amyand's hernia is even less frequent (0.07-0.13%), with an abdominal sepsis mortality of 15-30%<sup>(3)</sup>.

It is often a coincidental intraoperative finding, both in cases of appendicitis –since it looks like a complicated hernia– and in cases of scheduled hernia repair, with the patient being asymptomatic. In a 60-case review, only one case was diagnosed with Amyand's hernia preoperatively<sup>(4)</sup>.

Left Amyand's hernia is even less frequent as the cecal appendix is usually located in the right iliac fossa and it tends to be associated with intestinal malrotation, *situs*



**Figure 3.** Cecal appendix inside the left hernia sac (Amyand's hernia).

*inversus*, or mobile cecum, which change the laterality of the intra-abdominal viscera<sup>(1,2)</sup>.

In the case described, the left Amyand's hernia is related to the intestinal malrotation associated with the omphalocele. The latter is associated with congenital abnormalities in up to 70-75% of cases, such as Beckwith-Wiedemann's syndrome, cardiac and diaphragmatic malformations as a result of superior fold fusion disorders, bladder exstrophy and sacrococcygeal region defects owing to inferior fold fusion disorders, intestinal development and mesenteric abnormalities (intestinal malrotation, failed mesenteric implantation, intestinal atresia, and ischemic areas)<sup>(5,6)</sup>, and inguinal hernias, as it is the case here.

**Table 1. Amyand’s hernia classification and treatment recommendation according to Losanoff and Basson<sup>(7)</sup>.**

|          | <i>Findings</i>  | <i>Treatment</i>   |
|----------|--|--|
| Type I   | Normal appendix  | Prophylactic appendectomy<br>Herniotomy  |
| Type II  | Acute appendicitis, without peritonitis                                    | Appendectomy through herniotomy incision<br>Herniotomy                                     |
| Type III | Acute appendicitis with peritonitis or contamination of the abdominal wall | Appendectomy through laparotomy<br>Herniotomy  |
| Type IV  | Acute appendicitis associated with other abdominal pathology               | Appendectomy through laparotomy<br>Herniotomy<br>Control of associated abdominal pathology |

Omphalocele repair was performed once the newborn had been hemodynamically stabilized and other potentially life-threatening associated malformations had been ruled out. The technique and protocol used should be determined by newborn condition, coexistence of other severe malformations, condition of the peritoneal membrane covering the viscera (intact or broken), and defect size<sup>(5,6)</sup>.

In infants, inguinal hernia should be repaired at diagnosis, regardless of age, owing to the high risk of incarceration.

Right Amyand’s hernias are classified depending on the presence or absence of swollen appendix inside the hernia as established by Losanoff-Basson’s indications, which also provide with treatment recommendations according to intraoperative findings<sup>(7)</sup> (Table 1).

In left Amyand’s hernias, prophylactic appendectomy is systematically recommended to avoid future diagnostic

delays and mistakes in case of acute appendicitis, owing to the infrequent appendix location<sup>(7)</sup>.

## CONCLUSIONS

Left Amyand’s hernia is very rare and typically associated with embryonic intestinal development abnormalities, such as *situs inversus*, intestinal malrotation (isolated or associated with other abnormalities such as gastroschisis or omphalocele), and mobile cecum.

Given that the abnormal left location of the appendix can pose significant challenges for future acute appendicitis diagnosis, prophylactic appendectomy during defect repair is recommended in left Amyand’s hernia, regardless of patient age.

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