A case of Amyand's hernia in a preterm infant

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Summary: The findings of a normal or inflamed appendix in a hernia sac is a rare finding intraoperatively. It is referred to as an Amyand's hernia, named after Claudius Amyand who first described it in 1735. We present a case of a 7-week-old male, born prematurely at 32 weeks' gestation, who presented clinically with an incarcerated right inguinal hernia. Intra-operative findings revealed an inflamed and ruptured appendix in the hernia sac. An appendectomy and herniotomy was performed with good results.

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Introduction

Amyand's hernia is a term used to describe a hernia containing the vermiform appendix. It was so named after Claudius Amyand who described the diagnosis in 1735, after he successfully performed an appendectomy on an 11-year-old boy with the above presentation. De Garengeot hernia is used to describe an appendix found in a femoral hernia. Preoperative diagnosis is not common, and the presentation is usually of an incarcerated inguinal hernia.

Case Report

We present a case of a 7-week-old male, born prematurely at 32 weeks' gestation, as a result of severe pre-eclampsia in the mother. The presenting symptoms were of a 2-day history of a right inguinal swelling, and a distended abdomen. On admission the infant was apyrexial, he had a pulse rate of 156 and appeared clinically well hydrated. The abdomen was not distended and there were no signs of peritonitis. Examination of the scrotum revealed a swollen right scrotum that was firm, erythematous, with an impression of tenderness. The scrotal mass was irreducible. The systemic examination was grossly normal. The clinical assessment made was that of an incarcerated inguinoscrotal hernia. The blood results showed a white cell count of 16.07, a haemoglobin of 7,7 g/dl, and a thrombocytosis of 550. The urea and electrolytes were within normal range, and the arterial blood gas revealed a normal lactate. The infant was subsequently taken to theatre for an incarcerated inguinal hernia. Intraoperatively the sac was found to contain an inflamed, perforated appendix (Figure 1 and Figure 2), with pus noted. An appendectomy and herniotomy was done with irrigation of the scrotal sac, and a pencil drain was left in situ. Broad spectrum antibiotic cover was commenced. The infant had an uneventful

postoperative course and was discharged on postoperative day 5. The histopathology report confirmed acute gangrenous appendicitis with associated peritonitis. At the two-week follow-up the infant was noted to be well and was discharged from the surgical unit.

Discussion

A hernia is defined as the protrusion of an organ or fascia through the wall of the containing cavity. The contents of an inguinal hernia usually include small bowel and omentum. The incidence of a normal appendix in an inguinal hernia is 1% and this drops further to 0,1% when looking at cases of inflamed or perforated appendices.^{1,4,9} An Amyand's hernia is therefore a relatively rare entity. It is also rare in neonates and infants^{1,6}; more common in males and occurs most commonly on the right side.⁴ It has, however, been described to occur on the left in various clinical situations where the appendix may be found on the left side (situs inversus, intestinal malrotation, a long appendix or a very mobile caecum).^{12,11}

The pathophysiology of Amyand's hernia and its relationship with appendicitis are not fully known. Whilst some authors suggest that it is an incidental finding, others attribute it to incarceration leading to decreased blood supply and subsequent necrosis.⁶ Preoperative manoeuvres to reduce the hernia have also been postulated as a cause.^{8,11,13} Neonatal appendicitis occurs in 0,1% of all infantile cases of appendicitis. An Amyand's hernia has been implicated as a cause for neonatal appendicitis in 25–30% of all cases reported in the literature,^{[6,13,14} however various other causes have also been described. Hirschsprung's disease, cystic fibrosis, meconium plug syndrome, necrotizing enterocolitis and vascular insufficiency (cardiac or respiratory disease) are documented risk factors for appendicitis.^{12,13} The incidence



Figure 1. Inflamed appendix in hernia sac

of appendicitis and Amyand's hernia appears to be higher in prematurity¹² which is further illustrated in our case.

The clinical presentation is often of an incarcerated or strangulated inguinal hernia,6 and the diagnosis is usually made intraoperatively (as seen in our case).^{1,9} Preoperative diagnosis is unusual and is often difficult; imaging with computed tomography scan can suggest the diagnosis.3 In most clinical scenarios, however, due to clinical findings of incarceration, the patient is often taken to theatre for emergency surgery. Imaging is therefore not done. The differential diagnosis may include a strangulated hernia, orchitis, inguinal lymphadenitis and epidydimitis.

Management requires urgent exploration of the inguinal region and is dependent on whether the appendix is inflamed or not.6 In our case an inguinal incision was used. Our patient was managed with an appendectomy via the hernia sac and a herniotomy was performed (as advocated for paediatric inguinal hernia repair).11 Laparotomy may be necessary if there is gross peritoneal contamination. Some authors advocate for laparoscopic exploration of an inguinal hernia in infants.^{5,13} Instances where this approach is useful is when the hernia has previously been incarcerated, or when manual reduction has been difficult.¹³ The advantage of laparoscopic surgery is the ability to inspect the contralateral side as well the incarcerated organ.^{5,13} Incidental appendicectomy in the case of a normal appendix is controversial as the appendix has been used for various reconstructive procedures.^{4,11} Due to its anatomical and structural features, the appendix has been used as a conduit for biliary, urethral, vaginal, oesophageal and voice reconstructive procedures.¹⁵



Figure 2. Inflamed and ruptured appendix

Conclusion

Amyand's hernia is a relatively rare occurrence in the paediatric population with the preoperative diagnosis often being challenging. The presentation is often that of an incarcerated inguinal hernia. Management is surgical and in children includes herniotomy with or without appendectomy depending on the intraoperative findings.

Requirements of authorship

All authors contributed to the drafting and critical revision of this case report, and approved the version to be published.

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31

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