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CASE REPORT

# Amyand's hernia in premature twins

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**Abstract** Amyand's hernia (AH) is the presence of a normal or inflamed vermiform appendix in an inguinal hernia sac. This condition is unusual in neonates and in infants, and it has not been described in premature twins. We present two 32-day-old biovular twins with a right AH, treated with sparing of the appendix and herniotomy. The correct management of AH is discussed via a brief review of the literature.

**Keywords** Amyand's hernia · Prematurity · Appendix · Inguinal hernia

## Introduction

An inguinal hernia classically contains in its sac the omentum, the small bowel or the bladder. However, rarely it is possible to find a Meckel's diverticulum (Littre's hernia), a portion of the circumference of the intestine (Richter's hernia) or an inflamed or normal vermiform appendix (Amyand's hernia, AH) [1].

In 1736, Claudius Amyand first described the presence of a perforated appendix in the inguinal hernia of an 11-year-old-boy, operated at St. George's Hospital, London, UK [2, 3]. He reported this case in the journal *Philosophical Transactions of the Royal Society*, commenting "it is easy to conceive that this operation was as painful to the patient as laborious to me" [4]. Since his description, this finding appears to be relatively common in adults and in children; however, it appears anecdotal in neonates and premature neonates. In fact, as reported, in the literature, nearly 20 cases have been described [5–7]. To our knowledge, there are no published cases describing the contemporary presence of AH in premature twins. Herein, we present this singular presentation in two biovular premature twins.

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## Case reports

B.A. and B.S., two 32-day-old premature twin males (35-week-gestation) were electively admitted to our Division of Paediatric Surgery with the diagnosis of right inguinal hernia. They were clinically well, feeding normally and had a weight of, respectively, 3.250 and 3.450 kg. The differing weights of the two babies appeared to be occasional, without clinical significance. The past medical history was negative for B.A., while B.S. had respiratory distress during the delivery (Apgar score: 1′6, 5′10) that needed respiratory resuscitation.

The physical examination revealed in both twins a swelling in the right groin with omolateral hydrocele. In both twins, the surgical exploration of the hernial sac revealed a normal uninflamed vermiform appendix within the sac. The appendix was reduced into the abdomen and the herniotomy completed with high ligation of the sac. The postoperative courses were uneventful.

## Discussion

The presence of an uninflamed appendix, as an occasional intraoperative finding in inguinal hernia surgery, occurs usually three times more often in children than adults because of the patency of the processus vaginalis [8, 9].

This association could be related to the presence of a congenital band extending from the appendix into the scrotum up to the right testis and the funnel-shaped tapering caecum of the neonate [9–11]. However, the real pathophysiology of AH remains uncertain. The most common presentation of AH is with acute appendicitis, probably due to the vulnerability of a vermiform appendix included in a hernial sac, with its stretching and consensual reduction of its blood supply [7]. This feature is usually confused with an obstructed or strangulated inguinal hernia, and, especially in the neonatal age, with torsion of the testis or with epididymo-orchitis. The preoperative diagnosis of AH is very difficult; however, it has been described in a case of a 3-month-old male with ultrasonographic diagnosis of sliding appendiceal inguinal hernia [12]. The presence or absence of inflammation of the appendix in AH determines its treatment. The literature is unclear, but we believe that an incidental appendectomy should be avoided in the case of a normal vermiform appendix. The normal appendix should be gently reduced into the abdomen, reducing the risk of oedema or haematomas [13]. The sparing of a normal appendix is, in our opinion, preferable, due to the importance of its lymphoid tissue, the documented complications of an appendectomy, the high risk of wound infection (3–11%) and the potential for the later utility of an appendix [14, 15]. In particular, the appendix could be used for a urinary diversion, for biliary tract reconstruction and for anterograde bowel enemas.

## Conclusion

Amyand's hernia (AH) in the paediatric age remains anecdotal, especially in premature neonates. The universal treatment of AH is herniotomy; however, the literature is controversial regarding the management of the appendix. We suggest the sparing of an uninflamed appendix due to its potential for later utility.

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