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CASE REPORT

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Amyand's hernia presenting as neonatal testicular ischaemia

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Abstract Testicular ischaemia presenting in the neonatal period is most often attributable to neonatal torsion. We present an unusual case of a male neonate who presented with acute appendicitis within a patent processus vaginalis, causing cord compression and consequent testicular ischaemia.

Keywords Amyand's hernia · Neonatal appendicitis · Neonatal testicular torsion

Introduction

Established testicular infarction is invariably the operative finding in neonatal testicular torsion diagnosed at birth—a fact that sways some surgeons to delay immediate surgical intervention. Torsion developing in the neonatal period appears in some reports to have a higher salvage rate but the risk of metachronous torsion is such, however, that most will recommend contralateral fixation, if not immediately, then shortly thereafter. There is, however, no evidence base to, nor consensus on best practice. This report emphasises the fact that despite strong clinical suspicion of neonatal testicular ischaemia not all cases are due to extra-vaginal torsion. When dealing with possible neonatal testicular ischaemia, full consideration should be given to alternate diagnoses, which may require urgent intervention rather than elective surgical management.

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Case report

A 10-day-old 38-week-gestation male presented with a 2-day history of scrotal swelling and a hard right testis—possibly associated with tenderness. He was clinically well, feeding normally and was afebrile. Examination revealed the right hemi-scrotum to be hyperaemic and swollen. The spermatic cord was palpable in the right groin and slightly thickened.

Testicular and abdominal ultrasound suggested testicular ischaemia with a faint vascular signal within the right testis that lay in a large septated hydrocele. The left testicle was normal. The clinical impression was one of neonatal testicular torsion and in keeping with the delayed presentation it was felt the testicle was unlikely to be viable. Nonetheless, the decision was taken to explore the testes urgently and secure contralateral testicular fixation.

On exploration through a midline scrotal incision, serosanguinous fluid was aspirated from the right hemiscrotum revealing a testis that appeared pale but was otherwise well perfused and viable. Accordingly exploration of the groin was undertaken on suspicion of a covert inguinal hernia. The inguinal canal contained an oedematous and thickened spermatic cord with a patent processus vaginalis in which was incarcerated a gangrenous, perforated vermiform appendix (Fig. 1). Appendicectomy was undertaken through the internal ring as was laparoscopic evaluation of the abdomen to ensure no residual intra-abdominal sepsis. The right testicle was biopsied and fixed by placement in a Dartos pouch.

Culture of fluid taken from the processus vaginalis grew *Bacteroides fragilis* and Coliform species. Histological examination of the appendix subsequently confirmed acute inflammatory changes with infarction and the testicular biopsy confirmed haemorrhagic but otherwise normal appearances. The patient made an uneventful recovery and was discharged home on the third post-operative day.

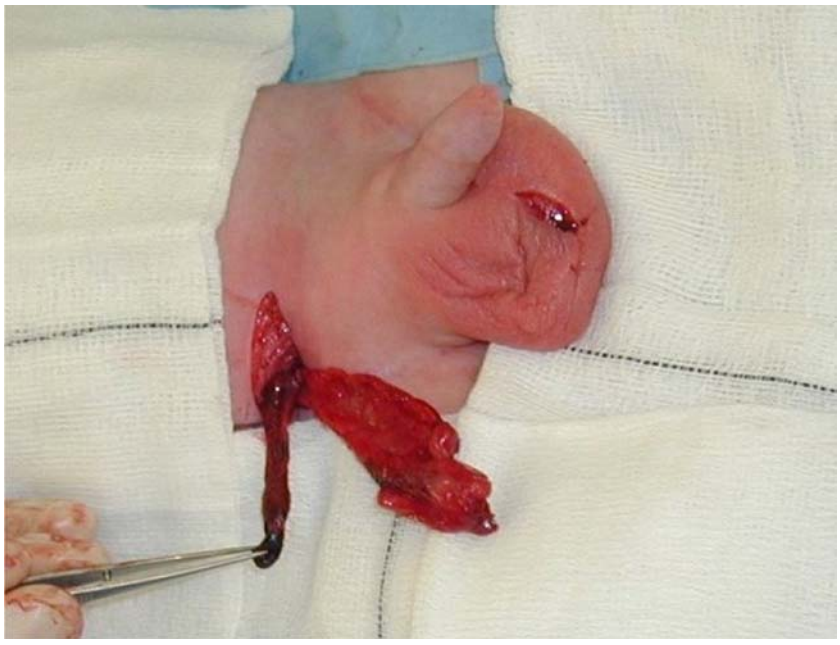

Fig. 1 The contents of the patent processus vaginalis. The infarcted appendix is displayed

Discussion

In 1735, Claudius Amyand performed the first appendicectomy on a perforated appendix within the inguinal hernia sac of an 11-year-old boy [1]. His name has now become eponymous with this rare condition. A normal appendix in an inguinal hernial sac is thought to occur three times more often in infants than adults due to persistent patency of the processus vaginalis [2]. Moreover, the predisposition has been associated with a congenital band extending from the appendix into the scrotum and attached to the right testis [3] and the funnel-shaped tapering caecum of the neonate, which allows the appendix to enter the processus vaginalis [4].

A review of all English language cases of neonatal appendicitis from 1901 to 2000 demonstrated that 32 of 128 documented cases (25%) lay within an inguinal hernial sac [5]. Since this time, two further cases in all languages have been reported of neonatal appendicitis within a hernia [6, 7]. The commonest presentation is that of an incarcerated inguinal hernia with the diagnosis being made during herniotomy. Testicular torsion has been considered as part of the differential diagnosis [8] but the earliest previous presentation with a pre-operative diagnosis of testicular torsion was 10 weeks old in an infant where the processus vaginalis was filled with pus and the appendix lay within the abdomen [9]. Our case represents an exceptionally rare event where the pre-operative diagnosis was of neonatal testicular torsion—a clinical diagnosis in part reinforced by ultrasound findings—but in retrospect the ischaemia was a consequence of pressure on the adjacent cord structures from the infarcted appendix within the confines of the processus vaginalis.

In the current case, the delayed presentation as neonatal testicular torsion may not have prompted urgent exploration by all, given the poor prospects of viability of the ipsilateral testis. The majority of perinatal testicular torsion is considered to be an intra-uterine event although up to 28% may occur post-natally [10]. If urgent surgical intervention is performed 40% of the latter group's testes may be salvageable if a normal neonatal examination has been recorded [11]. The risk of bilateral torsion and risk of anorchia if surgery is delayed also supports a need for urgent exploration [12]. This case highlights the need to consider alternate diagnoses in suspected neonatal testicular ischaemia and supports the practice of immediate surgical exploration.

References

1. Creese PG (1953) The first appendectomy. Surg Gynecol Obstet 97(5):643–652
2. Iuchtman M, Kirshon M, Feldman M (1999) Neonatal pyoscrotum and perforated appendicitis. J Perinatol 19(7):536–537
3. Oguzkurt P, Kayaselcuk F, Oz S et al (2001) Sliding appendiceal inguinal hernia with a congenital fibrovascular band connecting the appendix vermiformis to the right testis. Hernia 5(3):156–157
4. Firor HV, Myers HA (1964) Perforating appendicitis. Surgery 56:581–583
5. Karaman A, Cavusoglu YH, Karaman I et al (2003) Seven cases of neonatal appendicitis with a review of the English language literature of the last century. Pediatr Surg Int 19: 707–709
6. Martins JL, Peterlini FL, Martins ECS (2001) Neonatal acute appendicitis: a strangulated appendix in an incarcerated inguinal hernia. Pediatr Surg Int 17:644–645
7. Guelouz N, Rigourd V, Dommergues MA et al (2003) Appendicite aigue neonatale intra-herniaire. Arch Pediatr 10:1079–1082

8. Heydenrych JJ, Du Toit DF (1982) Unusual presentations of acute appendicitis in the neonate. South Afr Med J 62:1003–1005
9. Nagel P (1984) Scrotal swelling as the presenting symptom of acute perforated appendicitis in an infant. J Paediatr Surg 19:177–178
10. Pinto KJ, Noe HN, Jerkins GR (1997) Management of neonatal testicular torsion. J Urol 158(3):1196–1197
11. Sorensen MD, Galansky SH, Striegl AM et al (2003) Perinatal extravaginal torsion of the testis in the first month of life is a salvageable event. Urology 62(1):132–134
12. Olguner M, Akgur FM, Aktug T et al (2000) Bilateral asynchronous perinatal testicular torsion: a case report. J Pediatr Surg 35(9):1348–1349