Timely recognition of Amyand's hernia with appendicitis in infants

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Case report

Background: **Amyand's hernia with acute appendicitis is rare in infants and is mostly delayed in diagnosis and treatment, resulting in a high morbidity.**

Methods: **We presented two cases of infantile Amyand's hernia with acute appendicitis.**

Results: **Early surgical interventions were performed and both patients recovered without complication.**

Conclusion: **A practical strategy for this entity is to be aware of the rare disease and to perform early surgical exploration for suspected cases.**

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Key words: **Amyand's hernia; appendicitis; infant; morbidity**

Introduction

Amyand's hernia is an inguinal hernia with the appendix, whether normal or inflamed, in the hernial sac. It was first described in 1735 by Claudius Amyand, who performed a successful appendectomy on an 11-year-old boy who presented with an appendix in the hernial sac. Amyand's hernia with normal appendix is not rare and could be found incidentally during herniotomy, but Amyand's hernia with acute appendicitis is rare in children, especially in infants. Only a few cases of infantile Amyand's hernia with acute appendicitis were reported in the English literature and most of these cases were delayed in diagnosis and treatment with a high morbidity. Here we present two cases of infantile Amyand's hernia with acute appendicitis in which successful surgical interventions were performed without morbidity. To the best of our knowledge, case 1 is the first female infant case ever reported in detail.

Case report

Case 1

A 6-month-old female infant was admitted into our department with a 14-hour history of excessive crying associated with swelling and erythema in the right inguinal region. The infant did not present vomiting and had no history of inguinal hernia on either side. On physical examination, the infant was in a good general state and afebrile; the abdomen was flat and soft with hypoactive bowl sounds; an irreducible protrusion was found in the right inguinal region, tender to palpate. Routine blood tests disclosed mild leucocytosis. Ultrasound examination did not find ovary or intestine in the mass but suggested that the mass extended into the abdominal cavity through the internal ring. Surgical exploration was determined. Transverse incision was made in the lowest skin crease of the right inguinal region. The hernial sac was found extremely thickened, suppurate and fragile. On opening the hernial sac, the tip of appendix was found incarcerated within (Fig. A). The appendix was inflamed and adhered to the sac. Appendectomy was performed via the hernial sac and herniotomy was carried out after thorough saline irrigation. The postoperative course was uneventful and she was discharged on the 2nd postoperative day.

Case 2

A 15-day-old premature male (30-week-gestation) who had a history of right inguinal hernia was treated in the neonatal ward for pneumonia when a mass in the right inguinal region was found. No fever, vomiting or abdominal distention was presented. Ultrasound examination confirmed the bowel echo inside and the bowel component was reduced manually. But there was still a thickening palpated without clearly-

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defined margins, extending from the inguinal region towards the scrotum. One hour later, the bowel was incarcerated and manually reduced again. After the second reduction, the thickening still existed. Another 2 hours later when the bowel was incarcerated for a third time, the inguinal region was found to be tender and erythematous. The patient was subjected to surgical exploration. The hernial sac was found to be extremely thickened and ill-defined. On opening the sac, the appendix and cecum were found. The cecum was easily reduced but the appendix was tightly incarcerated in the distal hernial sac (Fig. B). The distal half of the appendix was obviously inflamed and there was a little pus in the distal sac. Appendectomy and herniotomy were performed via the same incision. The patient recovered uneventfully.

Discussion

Amyand's hernia with acute appendicitis in infants was mostly delayed in diagnosis and treatment. Although the general prognosis was good and no mortality was reported due to the localization of the inflammatory process, the delay in treatment resulted in a series of complications. The case reports in the English literature in the recent 30 years are presented in Table.[1-16] Eleven (64.7%) patients had their appendix perforated; at least 4 (23.5%) had scrotal abscess formation, 3 (17.6%) presented with peritonitis and 1 (15.9%) was in shock. Six (35.3%) patients required another abdominal incision to perform appendectomy. The delay was mainly ascribed to the doctors' unawareness of the rare disease. In none of the cases reported did the doctor ever think of the entity before operation.

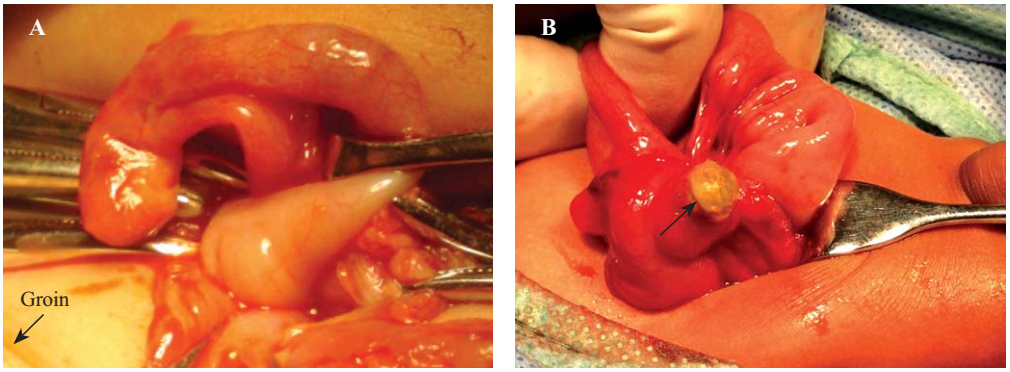

Fig. A: Female, 6-month-old. The appendix was incarcerated in the hernia sac and inflamed in the distal half; **B**: Male, 15-day-old. The appendix and cecum were found in the hernial sac. The distal half of the appendix was tightly incarcerated in the distal sac. The serosa of the cecum near the base of the appendix (arrow) was torn when opening the thickened and ill-defined hernial sac.

Table. Clinical information of the infants reported in English literature during the last 30 years

Author, publishing year	Sex	Mature/ premature	Age	Duration before operation	Preoperative diagnosis	Scrotal abscess/ peritonitis	Abdominal incision needed	Severity of appendicitis
Piedade, 2013[1]	Male	Mature	6 wk	15 d	Strangulated inguinal hernia with bowel perforation	Scrotal abscess	No	Gangrenous and perforated
Ergaz, 2013[2]	Male	Premature	30 d	-	Strangulated inguinal hernia	No	No	Perforated
Jain, 2012[3]	Male	Mature	1 y	9 m	Chronic scrotal sinus	Chronic scrotal sinus	No	Perforated
Khan, 2011[4]	Male	Mature	10 mon	10 h	Left testicular torsion	No	Yes	Inflamed
Rehman, 2010[5]	Male	Mature	8 wk	4 d	Swelling after reduction	No	No	Gangrenous
Park, 2010[6]	Male	Premature	33 d	13 d	Right epididymo-orchitis	Scrotal abscess	No	Inflamed*
Ngom, 2010[7]	Male	Mature	14 d	18 h	Strangulated inguinal hernia	No	No	Perforated
Livaditi, 2007[8]	Male	Premature	35 d	1 d	Strangulated inguinal hernia	Local peritonitis	Yes	Perforated
Livaditi, 2007[8]	Male	Premature	32 d	Short	Strangulated inguinal hernia	No	No	Inflamed
Milburn, 2006[9]	Male	Mature	10 d	2 d	Neonatal testicular torsion	Scrotal abscess	Yes	Gangrenous and perforated
Managoli, 2004[10]	Male	Mature	13 d	2 d	Peritonitis and shock	Local peritonitis	Yes	Perforated
Yazici 2003[11]	Male	-	8 mon	1 d	Elective herniotomy after reduction	No	No	Inflamed
Oguzkurt, 2001[12]	Male	Mature	9 mon	1 d	Swelling after reduction	No	No	Inflamed
Martins, 2001[13]	Male	Mature	4 d	-	Strangulated inguinal hernia	No	Yes	Perforated
Bannister, 2001[14]	Male	Premature	39 d	-	Strangulated inguinal hernia	No	No	Inflamed
Iuchtman, 1999[15]	Male	Mature	6 d	-	Strangulated inguinal hernia	No	No	Perforated
Halevy, 1988[16]	Male	-	20 d	-	-	Local peritonitis	Yes	Perforated and detached

*: Only the cases that were reported in detail were included; "-": no data.

The second reason for the delay was the difficulty in pre-operative diagnosis and determination for surgical exploration, especially in cases without the presence of bowel obstruction. When it occurred without bowel incarceration, the manifestation was nonspecific and it was frequently misdiagnosed as testicular torsion,[4] epididymo-orchitis[6] or soft tissue infection.[5] Ultrasound examination sometimes demonstrated a tubular structure extending into the inguinal canal and communicating with air-filled cecum, which was diagnostic for the disease; however, it could not be easily seen or not until the edema in soft tissue decreased in a relatively late stage.[6]

In our cases, both patients underwent early surgical exploration and the appendices were not perforated. The sign for the exploration in case 1 was the ultrasonographic finding that suggested the inguinal mass extended into the abdominal cavity. The sign in case 2 was the disproportionate swelling of the inguinal region after manual reduction, which was also presented in 2 cases reviewed.[5,12] Therefore, a practical strategy for this entity is to be aware of the rare disease and to perform early surgical exploration for suspected cases.

In conclusion, Amyand's hernia with acute appendicitis is rare in infants and has a high morbidity. A practical strategy for this entity is to be aware of the rare disease and to perform early surgical exploration for suspected cases.

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References

1 Piedade C, Reis Alves J. Amyand's hernia in a 6-week-old infant: a delayed diagnosis. Case Rep Pediatr 2013;2013:758171.
2 Ergaz Z, Simanovsky N, Vromen A, Meir K, Bar-Oz B. Amyand's hernia with perforated appendicitis in a premature infant. Eur J Pediatr 2013 [Epub ahead of print]
3 Jain P, Mishra A. Amyand's hernia presenting as chronic scrotal sinus. J Indian Assoc Pediatr Surg 2012;17:128-129.
4 Khan RA, Wahab S, Ghani I. Left-sided strangulated Amyand's hernia presenting as testicular torsion in an infant. Hernia 2011;15:83-84.
5 Rehman MR, Panteli C, Tsang T. Laparoscopic repair of Amyand's hernia in an 8-week-old infant. Hernia 2010;14:443-445.
6 Park J, Hemani M, Milla SS, Rivera R, Nadler E, Alukal JP. Incarcerated Amyand's hernia in a premature infant associated with circumcision: a case report and literature review. Hernia 2010;14:639-642.
7 Ngom G, Amadou I, Ibrahima KA, Mubake A, Ndour O, Ndoye M. Amyand's hernia with perforated appendix in a neonate. BMJ Case Rep 2010;2010.
8 Livaditi E, Mavridis G, Christopoulos-Geroulanos G. Amyand's hernia in premature neonates: report of two cases. Hernia 2007;11:547-549.
9 Milburn JA, Youngson GG. Amyand's hernia presenting as neonatal testicular ischaemia. Pediatr Surg Int 2006;22:390-392.
10 Managoli S, Chaturvedi P, Vilhekar KY, Gupta D, Ghosh S. Perforated acute appendicitis in a term neonate. Indian J Pediatr 2004;71:357-358.
11 Yazici M, Etensel B, Gürsoy H, Ozkisacik S, Erkus M, Aydin ON. Infantile Amyand's hernia. Pediatr Int 2003;45:595-596.
12 Oguzkurt P, Kayaselçuk F, Oz S, Arda IS, Oguzkurt L. Sliding appendiceal inguinal hernia with a congenital fibrovascular band connecting the appendix vermiformis to the right testis. Hernia 2001;5:156-157.
13 Martins JL, Peterlini FL, Martins EC. Neonatal acute appendicitis: a strangulated appendix in an incarcerated inguinal hernia. Pediatr Surg Int 2001;17:644-645.
14 Bannister SL, Wong AL, Leung AK. Acute appendicitis in an incarcerated inguinal hernia. J Natl Med Assoc 2001;93:487-489
15 Iuchtman M, Kirshon M, Feldman M. Neonatal pyoscrotum and perforated appendicitis. J Perinatol 1999;19:536-537.
16 Halevy A, Katz Y, Gross B. Auto-amputation of appendix in inguinal hernia. Clinical Pediatrics 1988;27:73.

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