CLINICAL CASE - TEST YOURSELF Pediatric imaging

Inguinoscrotal swelling in a neonate. Is it always a hernia?

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PART A

An 11-day old neonate presented to the emergency department due to right inguinoscrotal swelling. The clinical examination revealed an irreducible inguinoscrotal swelling with mild tenderness of the overlying skin. The right testis was not palpable probably due to local edema. The neonate was not irritable, could accept foods normally, and had bowel movements. Biochemical and hematologic laboratory parameters were normal. An ultrasound scan of the inguinoscrotal area was performed (Fig 1-3).

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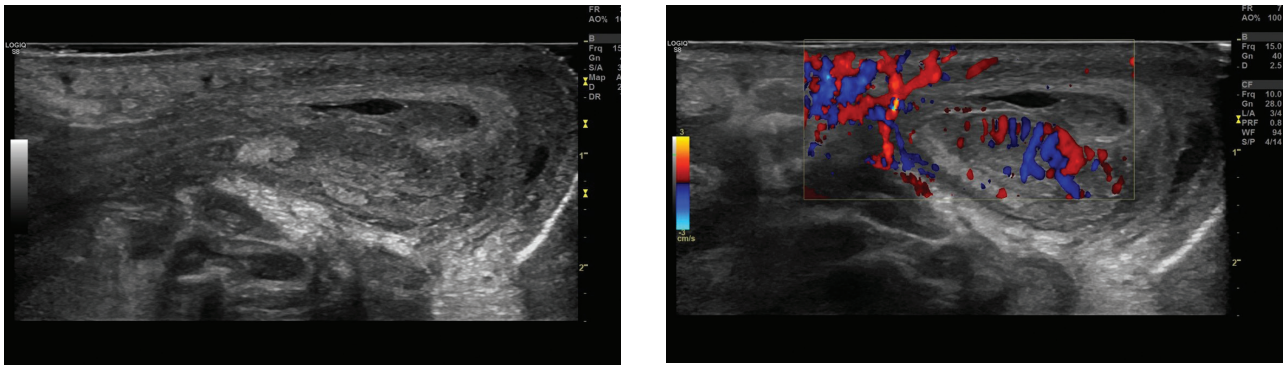

Fig. 1. *Longitudinal view of the inguinal canal and scrotum. a. Gray-scale US. b. Colour Doppler US.*

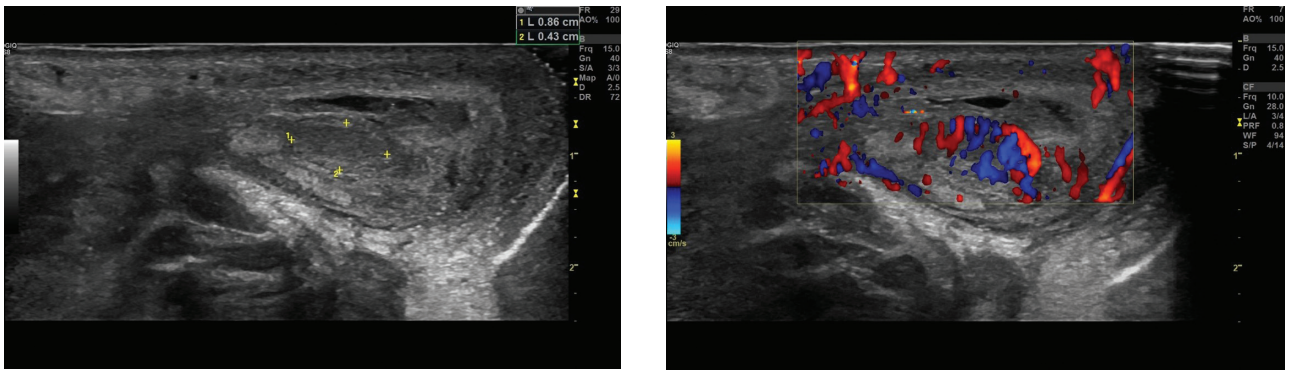

Fig. 2. *Longitudinal view of the scrotum. a. Gray-scale US. b. Colour Doppler US.*

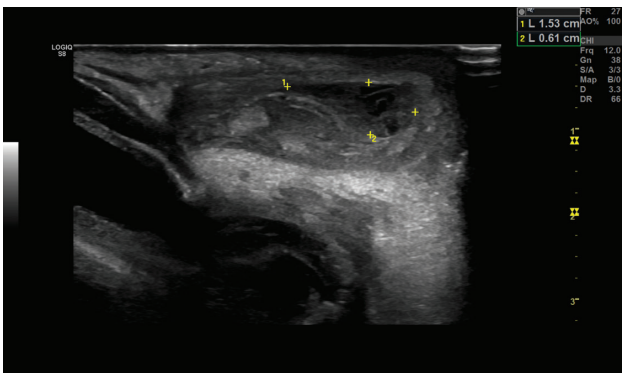

Fig. 3. *Gray-scale US. Longitudinal view of the inguinal canal and scrotum.*

PART B

Diagnosis: Scrotal suppurative appendicitis in a neonate (complicated Amyand's hernia-AH).

Ultrasonography (US) of the right scrotum revealed highly vascularized heterogeneous tissues extending into the inguinal canal and significant soft tissue edema of the scrotum (Fig. 1a, 1b). The testis was recognized (Fig. 2a), homogenous in morphology with increased internal blood flow (Fig. 2b) surrounded by turbid fluid (Fig. 3). There were no signs of abdominal extension, nor free fluid in the lower abdomen. The patient was taken to the operating room and perforated right intrascrotal appendicitis with abscess was found.

An inguinal hernia in a boy classically contains the omentum or/and the small bowel in its sac. However, unusual contents may be encountered, such as a Meckel's diverticulum (Littre's hernia), a portion of the circumference of the intestine (Richter's hernia), a normal or an inflamed vermiform appendix (Amyand's hernia) (Fig. 4) [1]. The presence of an uninflamed appendix, as an occasional intraoperative finding in inguinal hernia surgery, is usually three times more often in children than adults because of the patency of processus vaginalis and the mobile caecum. The reported incidence varies from 0.5% to 1% [2,3]. The proposed theories for this higher incidence in children are based on the presence of a congenital band that may extend through the inguinal canal connecting the appendix with the right testis. This interconnection, that can act as a guide of the appendix along with the funnel-shaped tapering caecum of the neonate are the two most possible pathogenetic factors. However, the real pathophysiology remains unclear [3].

The incidence of appendicitis within an inguinal hernia is even lower, representing 0.1% of all cases of acute appendicitis, but an accurate incidence in pediatric patients cannot be estimated because only a few case series have been reported in the literature [4]. Moreover, it was shown that in preterm and small for gestational age babies the appendix is more often inflamed when it is in the hernia sac as compared to the abdominal cavity [5].

The pathogenesis of appendicitis in the inguinal hernia has also been debated. Some authors suggest that the appendiceal inflammation is an incidental finding, while others argue that the incarceration of the appendix may provoke inflammation [3]. The latter support the theory that the appendix becomes entrapped due to contraction of the abdominal muscles against the sharp edge of the internal ring. This compromises the blood supply to the appendiceal wall, causes mucus accumulation, pressure necrosis, and inflammation [3,4].

Clinical presentation of neonates with inflamed AH varies considerably. Interestingly as in our case, the neonates usually present with inguinoscrotal swelling and erythema in the absence of laboratory findings. The narrow neck of the herniation sac seems to act as a protective barrier for the spread of inflammation in the abdominal cavity. Because of the limited peritoneal irrigation, the newborns are relatively active and continue to feed well despite ongoing appendicitis. Therefore, dehydration and circulatory stress are less frequently reported [5].

Although prompt intervention is being reported in neonates, the rate of appendiceal perforation remains high (50% in scrotal appendicitis) [5]. The thin appendiceal wall and the short omentum incapable of blocking a developing perforation are factors that can explain the higher complication rate in this age group. Other described complications in neonates include scrotal abscess (23.5%), peritonitis (17.6%), and enterocutaneous scrotal fistula (two case reports in the literature) [3,6]. Nevertheless, there is no mortality reported in the literature since 1975 [2,5]. Additionally, fulminant scrotal sepsis can theoretically induce thrombosis of testicular vessels and compromise testicular blood supply with consequent ischemia and necrosis. Fortunately, in a review of 24 neonates, it was concluded that testicular viability is not affected by the proximity of the perforated appendix [5].

Definite preoperative diagnosis presents a clinical challenge due to indistinct clinical signs and symptoms. Based on clinical criteria strangulated hernia, epididymo-orchitis, and testicular torsion were the 3 commonly improper diagnoses in neonates [5]. Combining high-frequency linear array transducers and Color Doppler, US can reliably disclose testicular torsion and epididymo-orchitis [7]. Okur et al and Almetaher et al reported the successful preoperative sonographic diag-

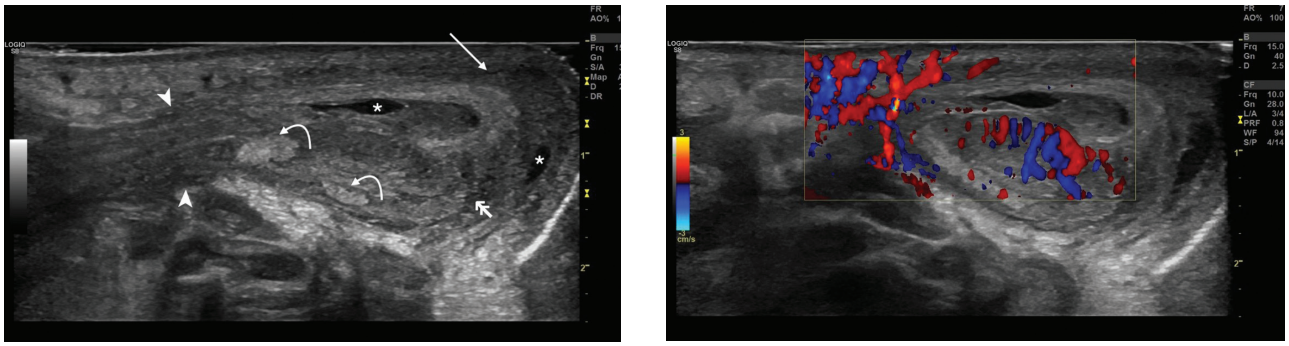

Fig. 1. *Complicated Amyand's hernia. a. Longitudinal gray-scale US of the inguinal canal and the scrotum demonstrates dilation of the deep inguinal canal (space between arrowheads). The scrotum contains heterogeneous tissues (double arrowhead), small fluid collections (asterisks), echogenic fat (curved arrows) consistent with intrascrotal fat necrosis and remarkable soft tissue edema (arrow). b. Same image with colour Doppler depicts markedly edematous and hyperemic tissues.*

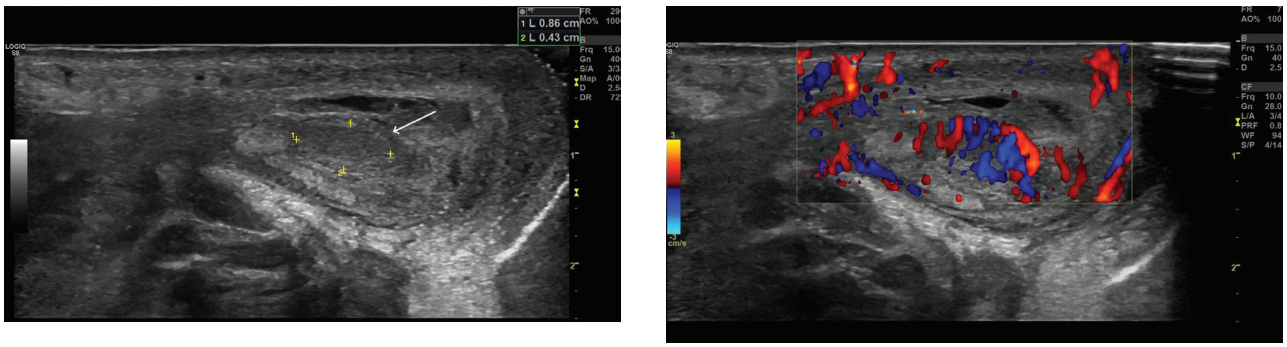

Fig. 2. *Complicated Amyand's hernia. Longitudinal view of the inguinal canal and the scrotum a. Gray-scale US. The testis (arrow) is recognized homogenous in morphology b. with increased internal blood flow on colour Doppler US.*

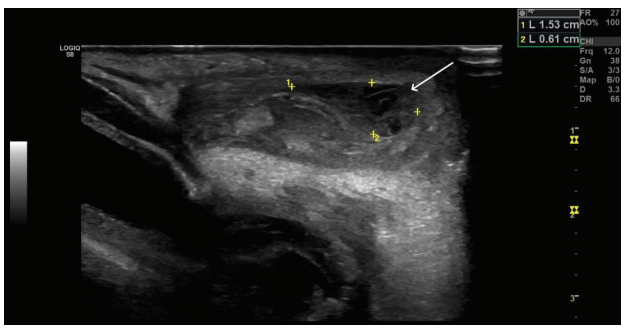

Fig. 3. *Complicated Amyand's hernia. Longitudinal view of the scrotum shows a heterogeneous area of mixed echogenicity with a small fluid collection (arrow) on its periphery.*

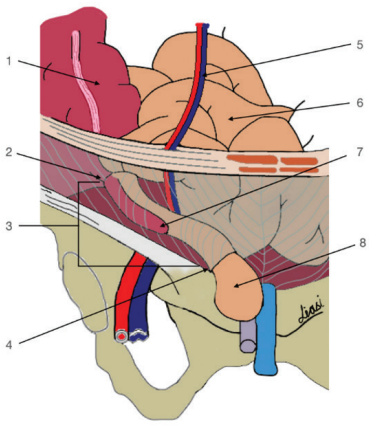

Fig. 4. *Amyand's hernia illustration. The appendix is depicted within the inguinal canal accompanying an indirect inguinal hernia which passes through both the deep and superficial inguinal rings (modified from source 1) .*
1 Large intestine; 2 Deep inguinal ring; 3 Inguinal canal; 4 Superficial inguinal ring; 5 Inferior epigastric vessels; 6 Small intestine; 7 Appendix; 8 Small intestine protruding through inguinal canal

nosis of AH in some of their patients (9 out of 12 and 3 out of 3 patients aged 2 months to 10 years and 15 days to 5 years respectively) [4,8]. In the aforementioned studies, the most common presenting symptom was that of irreducible hernia without symptoms of intestinal obstruction.

The sonographic diagnosis of a perforated AH is also challenging. In this scenario, as in our case, the diagnosis relies on the demonstration of secondary signs such as a heterogeneous area composed of internal echoes, membranes, and turbid fluid representing a scrotal abscess. A vascularized homogenous testis can usually be detected. Occasionally inflammatory fat and tissues extend into the inguinal canal, possibly communicating with air-filled caecum.

Treatment is that of urgent surgical exploration of the inguinal region. The surgical approach of AH in adults was described by Lossanoff and Basson [3]. In 2007 they proposed a classification system clarifying the appropriateness of mesh hernioplasty in the presence of infection (Table 1) [4]. However, there is still a lack of consensus on management protocol concerning pediatric patients presented with AH. The presence or absence of appendiceal inflammation determines the approach. In the presence of appendicitis, treatment involves an emergency appendectomy and hernia repair. On the contrary, in the case of incidentally found normal appendix in the hernia sac the performance of prophylactic appendectomy along with the hernia repair is not favored [3,9].

At last, a correct preoperative diagnosis requires awareness of this rare entity by the clinician. A radiologist informed about a possible AH should pay attention to the sonographic details increasing the possibility of preoperative diagnosis [5]. Finally, early diagnosis and treatment contribute to a fast and safe perioperative course with no morbidity [10].

Conclusion: Acute appendicitis presenting as a scrotal abscess is a very rare entity. It is an emergent condition needing source control over infection to prevent sepsis and should be considered in the differential diagnosis of acute scrotum. Proper management is dependent upon accurate recognition of the clinical signs and symptoms. We emphasize that the sonographic examination may exclude other pathologies mimicking acute scrotum. Finally, we highlight that it can identify and confirm a clinically suspected AH with possible complications thus leading to timely operative intervention. R

Conflict of interest

The authors declared no conflicts of interest.

Table 1

Types of AH	Features	Surgical management
Type 1	Normal appendix within the inguinal hernia	Reduction of appendix or appendectomy and mesh hernioplasty
Type 2	Acute appendicitis with no abdominal sepsis	Appendicectomy through the hernia and sutured hernioplasty
Type 3	Acute appendicitis with abdominal sepsis	Appendicectomy through laparotomy with sutured hernioplasty
Type 4	Acute appendicitis associated with related or unrelated abdominal pathology	Appendicectomy through hernia or laparotomy plus diagnostic workup

KEY WORDS | Ultrasonography; Inguinal Hernia; Perforated Appendicitis; Neonate

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