

Amyand hernia mimicking an acute scrotum in a newborn patient

Hernia de Amyand simulando un escroto agudo en un recién nacido

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ABSTRACT

Background: Acute scrotum in pediatrics is an emergency characterized by the sudden or progressive onset of pain, swelling, and redness in the scrotal area. It is crucial to promptly diagnose and treat the underlying cause to prevent serious complications.

Case presentation: A 19-day-old male newborn presented with a 4-day history of feeding difficulties, progressive irritability, and a single episode of fever peaking at 38.5°C. Physical examination revealed right scrotal swelling, non-reducible, with a non-palpable testis. A plain abdominal X-ray demonstrated the presence of gas at the inguinal region. Following surgical preparation, an inguinal incision was performed, leading to the diagnosis of a type 3 Amyand hernia. The postoperative course was uneventful, and the patient showed favorable recovery. **Conclusions:** An Amyand hernia can mimic an acute scrotum syndrome in pediatric patients. Depending on its intraoperative classification, treatment may be performed solely through an inguinal approach.

Keywords: Acute scrotum; Pediatrics; Inguinal hernia; Amyand's hernia

RESUMEN

Introducción: El escroto agudo en pediatría es una emergencia caracterizada por la aparición súbita o progresiva de dolor, inflamación y enrojecimiento en la zona escrotal. Es crucial diagnosticar y tratar rápidamente la causa subyacente para prevenir complicaciones graves. **Presentación del caso:** Recién nacido de 19 días de edad acude por cuatro días de dificultad para la alimentación, irritabilidad progresiva y un episodio único de fiebre de 38,5°C. En el examen físico se evidenció un aumento de volumen en el escroto derecho, no reducible, y con testículo no palpable. Se realizó una radiografía simple de abdomen y se encontró gas a nivel inguinal. Tras la preparación quirúrgica, se realizó una incisión inguinal, diagnosticándose una Hernia de Amyand tipo 3. La evolución postoperatoria fue favorable. **Conclusiones:** La hernia de Amyand puede simular un síndrome de escroto agudo en pediatría. Dependiendo de su clasificación intraoperatoria, el tratamiento puede realizarse solamente mediante abordaje inguinal.

Palabras clave: Escroto agudo; Pediatría; Hernia inguinal; Hernia de Amyand

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INTRODUCCIÓN

Numerous conditions may lead to acute scrotum syndrome in pediatric patients; however, acute appendicitis is not typically regarded as a contributing cause.

Scrotal swelling in a newborn can be the result of conditions related to the patent processus vaginalis such as hydrocele, spermatic cord cyst or inguinoscrotal hernia, and/or testicular problems like a mass or the torsion of the gonad. Exceptionally, prenatal or perinatal bowel perforation may also present with scrotal redness and inflammation due to meconium passage through a patent processus vaginalis.^{1,2} Acute scrotum is a clinical syndrome characterized by the sudden onset of pain, swelling and/or redness of the scrotum or its contents. The pain may be truly sudden and severe or may have been progressing over a few days, depending on the etiology.² It represents an urological emergency that requires prompt evaluation and management to prevent complications, including testicular loss or damage.

The challenge to the pediatric surgeon is to differentiate conditions that require immediate surgical exploration from those that can be managed nonoperatively.

CASE PRESENTATION

A 19-day-old male newborn, from an uncomplicated vaginal delivery at 39.3 weeks of gestation, with no significant pre or perinatal history, was referred to the hospital by his primary care pediatrician due to 4 days of difficulty feeding, progressive and marked irritability, and a single episode of fever of 38.5°C.

On physical examination, upon admission to the Neonatology unit, right scrotal swelling with redness was noticed, with effacement of the scrotal folds, tenderness on palpation, and the testicle could not be palpated. The temperature was 38.7°C. The rest of the physical examination was unremarkable.

A consultation with the on-call pediatric surgeon was requested, with the preliminary diagnosis of acute scrotum. Upon being evaluated by the pediatric surgeon, a calmed newborn was described, who was sleeping at the time (Dipyrone had been administered for pain relief and to lower the temperature). The abdomen was slightly distended on inspection, diffusely tender on palpation, with decreased bowel sounds on auscultation.

Right inguinal region: a tubular shaped swelling was observed, which seemed to extend into the scrotum; the skin was tense with no color changes.

Right scrotum: significantly enlarged, reddened, with tense and smooth skin, and very tender when palpated. The right

testicle could not be palpated.

Left inguinal-scrotal region: no abnormalities. Left testicle was of normal size for his age, placed in a normal position, and not painful.

Blood test results showed a white blood cell count of 16.3 k/mm³, C reactive protein level of 8.0 mg/dl. There were no other significant abnormalities in his blood test results. Since an emergency ultrasound was not available, a plain abdominal film was ordered, including both inguino-scrotal areas. Imaging reveals dilated bowel loops with mural edema and the presence of gas in the right inguinal region, suggesting the presence of an inguinal hernia (Figure 1).

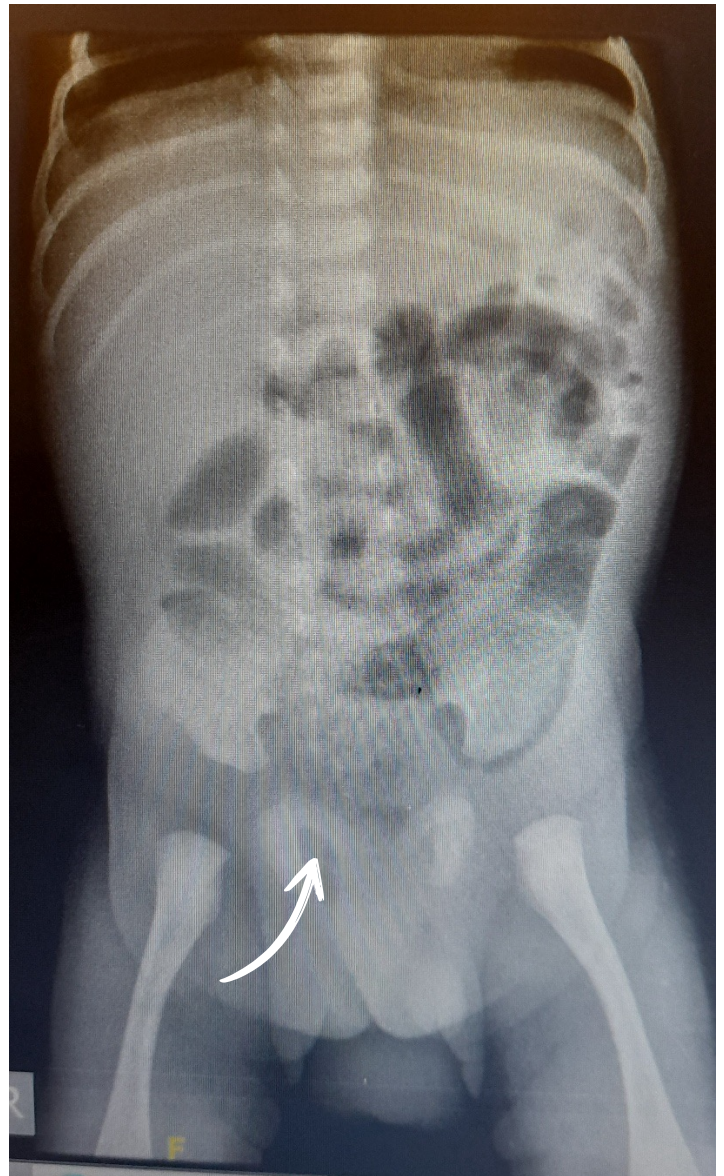


Figure 1. Plain radiography showing an image suggestive of gas in the right inguinal region (arrow).

The patient was scheduled for emergency surgery with the diagnosis of a right incarcerated inguinal-scrotal hernia; antimicrobial treatment with Ampicillin plus Sulbactam was

initiated according to the hospital protocol.

Surgical procedure

An oblique inguinosotomy was performed, deepened by layers until the external oblique muscle fascia was identified, which was hardened and edematous throughout its extension. A thickened, edematous spermatic cord was found, which was difficult to separate and free from the surrounding structures. The hernia sac was identified through blunt dissection, and it was opened between clamps, releasing purulent fluid from inside. A culture swab was taken and antibiotic sensitivity testing was also requested.

Upon opening the hernia sac proximally and distally, fibrin and a grayish structure adhered to the inner wall of the sac were observed, resembling a tubular shape. The structure was dissected and completely separated from the hernia sac, revealing a gangrenous cecal appendix along its entire length, including the mesoappendix. (Figures 2 and 3) The deep inguinal ring was opened, allowing to reach the base of the appendix, which had good color and aspect. An appendectomy was performed with double ligation of the appendicular base using 3/0 Vicryl. Subsequently, pus was aspirated from the pouch of Douglas, and the right and left paracolic gutters. The peritoneal cavity was irrigated with a warm 0.9% saline solution.



Figure 3. Completely mobilized gangrenous cecal appendix.

Despite the wide tissue inflammation, a right inguinal herniorrhaphy could be performed, preserving the spermatic cord elements. High ligation of the proximal end of the sac was done with 3/0 Vicryl, and the distal end was opened, releasing pus from the scrotum. The vaginal tunic was very edematous, showing an enlarged light blue testicle. The testis was immediately covered with gauze soaked in warm saline solution, and within five minutes, it had regained its normal pearly white appearance. The deep inguinal ring was partially closed with 3/0 Vicryl suture. Layered closure was performed down to the skin with 3/0 Vicryl and 4/0 Monocryl (Figure 4).



Figure 2. Grayish tubular structure found inside the hernia sac.



Figure 4. Oblique right inguinal incision after closure.

Postoperative course

During the surgery, the first dose of Gentamicin and Metronidazole was administered, and the treatment continued for 7 days along with Ampicillin.

The culture swab result from the peritoneal fluid was ready 3 days after surgery, and it showed polymicrobial flora. The antibiotic treatment continued with the same agents since the isolated bacteria were sensitive to them.

Oral feeding with breast milk was progressively resumed 48 hours after surgery, and the postoperative evolution was favorable. The patient was discharged from the hospital on the 8th day after surgery.

DISCUSSION

The widely accepted definition of Amyand's hernia is the presence of the appendix in the inguinal hernia sac, with or without inflammatory changes. It is named after Dr. Claudius Amyand, a French doctor and surgeon of the king of England, best known for performing the world's first successful appendectomy at St. George's Hospital in London on December 6th, 1735. The patient, Hanvil Anderson, was an 11-year-old boy who suffered from an inguinal hernia and a fecal fistula discharging in the groin. At operation, the appendix was found in the hernia sac and the fistula was traced to a perforation of the appendix by a pin. Amyand excised the appendix in an operation lasting almost half an hour.²⁻⁴ The case was reported in the Philosophical Transactions of the Royal Society in 1736, in which Amyand comments: "it is easy to conceive that this operation was as painful to the patient as laborious to me". Apparently, young Hanvil endured this ordeal with fortitude. He recovered, was cured of the faecal fistula but, not surprisingly, the hernia recurred.³

Amyand's hernia has been reported in a wide range of age groups, from newborns to the elderly. Its incidence is relatively low as it accounts for 0.19-1.7% of all inguinal hernias. The occurrence of appendicitis within Amyand's hernia is even less common, with its prevalence being estimated to be between 0.07% and 0.13%, with appendiceal perforation occurring in only 0.1% of those cases^{1,4-6}. Amyand's hernia complicated by appendicitis is reported to account for 0.1% of all cases of appendicitis^{2,4}. The ages of patients reported to have Amyand's hernia vary widely, from 3 weeks to 88 years. The condition is approximately three times more common in children than in adults due to the higher prevalence of a patent processus vaginalis in children.^{2,6-10}

Concomitant Amyand inguinal hernia with acute appendicitis is more common in children, mostly affecting males, and in most cases, it is located on the right side because the anatomically normal position of the appendix.^{1,2,4,5} However, there have been reported cases in which the condition

occurs on the left side due to excessive mobility, intestinal malrotation, a floating cecum, or *situs inversus*.^{2,6,9,10}

The exact mechanism by which appendicitis occurs in the inguinal hernia sac is not fully understood. However, several proposed mechanisms include: ^{1,2,6,8,11}

1. Inflammation due to incarceration.
2. Adhesions were formed as the appendix remained within the hernia sac for a long time, increasing the risk of injury.
3. Increased intra-abdominal pressure resulting from the contraction of abdominal muscles, leading to the compression and functional obstruction of the escaped appendix.
4. Swelling of the escaped appendix due to venous stasis, causing microcirculatory disturbances in the wall of the tubular organ, which can promote bacterial overgrowth and translocation.

Edan OA¹¹ reported a patient exhibiting a fibrous connection between the tip of the appendix and the testis. They believed that the fibrous connection guided the passage of the vermiform appendix into the hernial sac with the assistance of the patent processus vaginalis. In our patient, the cecal appendix was adhered to the inner wall of the hernia sac, which was relatively easily separated by blunt dissection. We cannot determine whether the appendix had any previous adhesion to the wall of the hernia sac, or if adhesions occurred because of the inflammatory appendiceal process.

This condition is usually asymptomatic, discovering the appendix without inflammatory changes during inguinal hernia repair. When symptoms occur, they are related to strangulated hernia rather than acute appendicitis, including groin pain, acute irreducibility, and sometimes nausea and vomiting. The local physical examination shows swelling of the groin, tenderness, redness, or even local necrosis in neglected forms.^{2,6,9-11}

Our patient presented with a single episode of fever on the day of admission and a history of irritability and difficulty feeding. The possibility of a complicated right inguinal hernia was greater than any other condition related to the testicle or epididymis. However, the inguinal region and the right scrotum, although slightly larger compared to the contralateral side, showed a difference that was not very significant. The diagnosis of Amyand's hernia was never suspected preoperatively.

Imaging studies are of great value in diagnosing Amyand's hernia, as it can be challenging to differentiate this condition from other diseases based only on clinical findings, especially in pediatric population.^{1,2,6} Plain abdominal X-ray, inguino-scrotal ultrasonography and CT scan are very useful for the

diagnosis of testicular problems, inguinal hernias, whether complicated or not, including Amyand's hernia.^{4-6,8,10} In our patient, the plain abdominal X-ray showed the presence of gas at the inguinal region, that helped us decide to proceed with the emergency surgical treatment.

Amyand's hernia treatment may vary depending on whether the appendix is complicated or not in the hernia sac, the presence of complications and/or other pathologies, which was described by Lossanof JE, *et al.*¹² The condition is classified into 4 types.

1. Type 1: Cecal appendix in the hernial sac without appendicitis. Treatment: Reduction of the appendix, or incidental appendectomy (controversial), and hernia repair.
2. Type 2: When there is appendicitis, but it is confined within the hernia sac. Treatment: Appendectomy through the inguotomy and hernia repair.
3. Type 3: Acute appendicitis complicated with peritonitis or abdominal wall contamination. Treatment: Appendectomy through laparotomy, cavity lavage and drainage, high orchiectomy, fasciotomy, and no prosthetic hernia repair
4. Type 4: When acute appendicitis is associated with another abdominal pathology, such as mucous cystadenoma or cancer in the appendix. Treatment: Appendectomy through laparotomy, no prosthetic hernia repair, and control of associated abdominal pathology.

There are publications advocating appendectomy in Amyand's hernia with a non-inflamed appendix to eliminate the risk of appendicitis later, especially those located on the left side.^{5-7,13} However, many surgeons do not perform prophylactic appendectomy in pediatric patients due to insufficient data, the risk of infection during a non-contaminated surgery, and the appendix can be used in other procedures if necessary.^{1,2,4,6,7}

The perforated appendix, in the context of Amyand's hernia, is more commonly observed in later childhood, with very few cases reported in the neonatal period. The presence of a narrow neck of the hernial sac, coupled with a small diameter impacted appendix will limit the peritonitis at the saccular level preventing stool leakage from the perforated appendix into the peritoneal cavity. In our patient, the deep inguinal ring was slightly dilated, allowing pus to pass into the peritoneal cavity and thus causing peritonitis, which was successfully treated through the inguinal approach.^{6,11}

According to the aforementioned classification, our patient suffered from Type 3 Amyand's hernia. For this group of patients, Losanoff JE, *et al.*, recommend that the treatment can include not only appendectomy and hernia repair but also more extensive surgical procedures, mainly in adults, such

as high orchiectomy, right hemicolectomy, and debridement for necrotizing fascitis, depending on the individual case.^{8,13} In this case, in addition to appendectomy, the surgical procedure included peritoneal cavity lavage with suction and inguinal hernia repair.

In a systematic review of appendicitis and its associated mortality and morbidity in infants up to 3 months of age in 2023,¹⁴ the authors reported fatal outcome only in patients with abdominal appendicitis and none among patients with herniated appendicitis; and morbidity was reported in only two patients out of 58 with herniated appendicitis. These findings may be attributed to two important factors; first, a local irreducible swelling as opposed to general abdominal symptoms allows for timely recognition and prompt treatment, more so as the differential diagnosis includes testicular torsion and incarcerated small bowel, both requiring immediate surgical treatment. Secondly, although several patients with Amyand's hernia presented with general symptoms and sepsis, the relatively anatomically contained feature of an Amyand's hernia may be a protective factor against poor outcome. Nevertheless, Ahmed HO, *et al.*,¹⁵ in a five-year study, described that once the appendix becomes inflamed or perforated within the hernial sac, it poses a 14-30% mortality rate, compared to a 0.5-5% mortality rate for an inflamed or perforated appendix in its normal anatomical site. Despite our patient presenting with generalized peritonitis, the postoperative outcome was favorable without any further complications.

CONCLUSIONS

Amyand's hernia is a rare condition mimicking an acute scrotum syndrome in pediatric patients, especially in neonates. Preoperative diagnosis is very difficult to establish, although the use of plain abdominal X-rays can be extremely helpful. Surgical treatment involves performing an appendectomy through the inguinal approach and repairing the hernia in the simplest cases, while in patients with generalized peritonitis, may also require peritoneal lavage and drainage. Postoperative outcome primarily depends on the inflammatory state of the appendix, the patient's clinical condition, antimicrobial treatment, and the availability of neonatal intensive care unit if needed.

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Authorship

- **YRA:** Conceptualization, data curation, research, methodology, visualization, writing (original draft), review and edition.
- **SDP:** Supervision, validation, review and editing.

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