

Acute perforated appendicitis with pre-surgical diagnosis and bilateral inguinal hernia in preterm infants

Apendicitis aguda perforada con diagnóstico prequirúrgico y hernia inguinal bilateral en recién nacido de pretérmino

Hernán Villalón U.^{a,b,c}, Angélica B. Patiño^d, Carolina Pérez S.^e,
Stefan Hosiasson S.^{a,c}, Mauricio Pinto C.^{a,c}, Patricio Varela^f

^aNeonatal Intensive Care Unit. Clínica Las Condes. Santiago, Chile.

^bUniversidad de Chile. Santiago, Chile.

^cDepartment of Paediatrics. Universidad Finis Terrae. Santiago, Chile.

^dIntern of Paediatrics. Universidad Finis Terrae. Santiago, Chile.

^eDepartment of Radiology. Clínica Las Condes. Santiago, Chile.

^fDepartment of Child Surgery. Clínica Las Condes. Santiago, Chile.

Received: December 3, 2019; Approved: Jun 28, 2020

What do we know about the subject matter of this study?

Appendicitis in newborns is very rare with high mortality due to the difficult diagnosis and late surgery, among other factors.

What does this study contribute to what is already known?

Increased understanding, improved resolution of ultrasound equipment, and trained specialists can significantly contribute to an early pre-surgical diagnosis that changes the prognosis.

Abstract

Acute neonatal appendicitis is a rare pathology, with few reports in the last 30 years. Since its clinical presentation and imaging studies are non-specific, most cases are diagnosed during a surgical procedure. **Objective:** To describe a neonatal case of acute perforated appendicitis associated with later appearance of inguinal hernia, with pre-surgical diagnosis and treated through laparoscopy. **Clinical Case:** A 17-day-old preterm female newborn presented with fever, irritability, and increased milk intolerance. Physical examination showed abdominal distention, tenderness and both, abdominal X-ray and ultrasound showed compatible images with acute perforated appendicitis. Once the diagnosis was confirmed, we performed an appendicectomy through laparoscopy. Two weeks later, during an outpatient visit, we observed a bilateral inguinal hernia which was confirmed by ultrasound, and then it was surgically corrected. **Conclusion:** Acute appendicitis should be considered within the differential diagnosis in neonates with acute abdominal symptoms, mainly in premature infants. In this case, ultrasound scan allowed us to make the pre-operative diagnosis. The possible association with inguinal hernias should motivate to examine inguinal ducts during imaging assessment and surgical procedure.

Keywords:

Acute Appendicitis;
Newborn;
Premature;
Acute Neonatal
Abdomen;
Neonatal Inguinal
Hernia;
Ultrasound;
Pre-Surgical Diagnosis

Correspondence:

Hernán Villalón

hvillalon@clinalascondes.cl

How to cite this article: Rev Chil Pediatr. 2020;91(6):936-940. DOI: 10.32641/rchped.v91i6.1538

Introduction

Neonatal appendicitis is a very rare pathology, with less than 50 cases reported in the last 30 years^{1,2,3}. In most of the reported cases, neonatal appendicitis was associated with some other pathology such as Hirschsprung's disease, meconium ileus, and uni- or bilateral inguinal hernia, among others⁴. The risk factors for this entity are related to conditions that cause hypoxia-ischemia¹, which could explain the pathophysiology of this picture.

Usually, the symptoms are nonspecific, predominating abdominal distension and vomiting, and, in most cases, there are symptoms characteristic of necrotizing enterocolitis². Ultrasound can be useful for diagnosis but has limitations³. In general, it is a pathology that quickly progresses to perforation and peritonitis², since it is usually diagnosed late and, most of the time, it appears as a surgical finding in a picture compatible with acute abdomen, and its treatment is surgical resection¹. Due to the infrequency of this diagnosis and its importance in the newborn, we present a clinical case of perforated acute appendicitis in a premature patient, with pre-surgical radiological diagnosis and laparoscopic resolution, associated with the subsequent development of an inguinal hernia.

Clinical Case

Female preterm newborn (34 weeks), twin I, child of a primiparous mother. Her mother had premature labor refractory to Atosiban, therefore a C-section with uncomplicated cephalic extraction was performed. She was born weighing 2,075 gr, length 43.5 cm, Apgar score 8-8, and normal physical examination. She was admitted to the Neonatal Intensive Care Unit, where a sample was taken for blood culture and then started combination antibiotic therapy. Within the first hours, the newborn presented respiratory distress managed with CPAP for 8 hours and then only with oxygen for 48 hours. Feeding was started at 24 hours of age, with good tolerance to the progressive increase. On the 3rd day of life, we decided to withdraw antibiotics because of negative blood cultures, two normal blood counts, and C-reactive protein tests.

At 17 days of age, she presented fever, irritability, and bilious gastric residuals. On physical examination, a soft but distended and tender abdomen stood out, with bowel sounds. A plain abdominal x-ray showed a pathological distribution of intestinal gas, with little representation in flank and right iliac fossa and the presence of isolated confluent gas bubbles suggesting extra-intestinal air (figure 1).

In the abdominal ultrasound, there was signifi-

cantly less gas in intestinal loops on the right side, with decreased peristalsis and thickened walls, along with increased echogenicity of the lateral and posterior fat layer in flank and right iliac fossa. There was scarce free fluid of echogenic aspect in the right iliac fossa (figure 2a). We partially visualized a 6-mm diameter appendix, in retrocecal position, thickened, ending in a poorly delimited area of heterogeneous hypoechoic and extraluminal mass with confluent gas bubbles, suggestive of perforation (figure 2b). With these findings, a probable complicated acute appendicitis was proposed. Inguinal canals were not described.

Among laboratory tests, a blood count with leukocytosis (24,700 per mm³) and C-reactive protein 54 mg/l stand out. Combination antibiotic therapy was started (Ampicillin, Amikacin, and Metronidazole). An exploratory laparoscopy showed a perforated appendix and free-flowing purulent fluid, therefore an appendectomy and cavity lavage were performed without exploring the inguinal ducts.

A biopsy confirmed acute perforated appendicitis without malignancy. In peritoneal culture, an ampicillin-sensitive *Enterococcus* strain was identified. After completing 7 days with antibiotics and presenting a good outcome, the patient was discharged.

Two weeks later, in outpatient care, she showed increased inguinal volume that could not be reduced. Ultrasound showed a bilateral hernia with left incarceration.

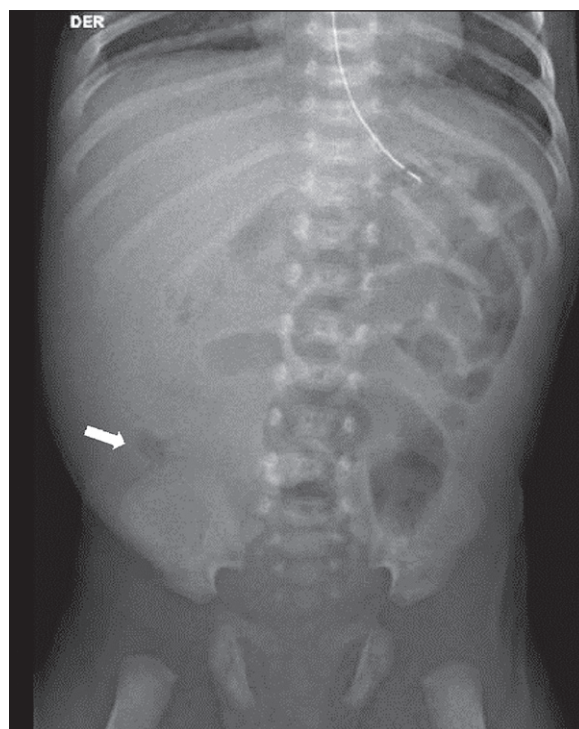


Figure 1. Plain X-ray of abdomen. It shows pathological air distribution and confluent bubbles in right lower quadrant suggesting extraintestinal air (indicated with arrow).

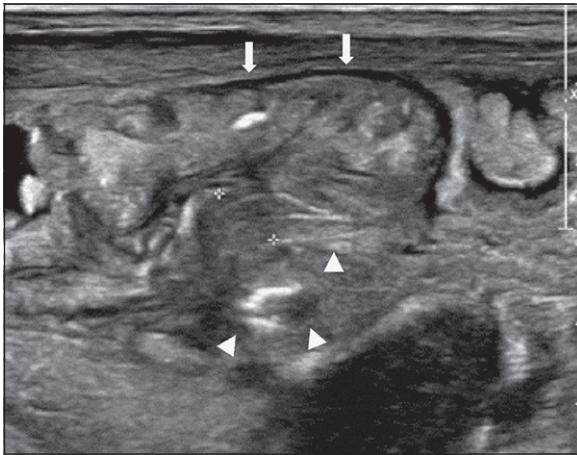


Figure 2a. Abdominal ultrasound scan: it shows thickening of intestinal loops and greater echogenicity in the flank adipose tissue and right iliac fossa.

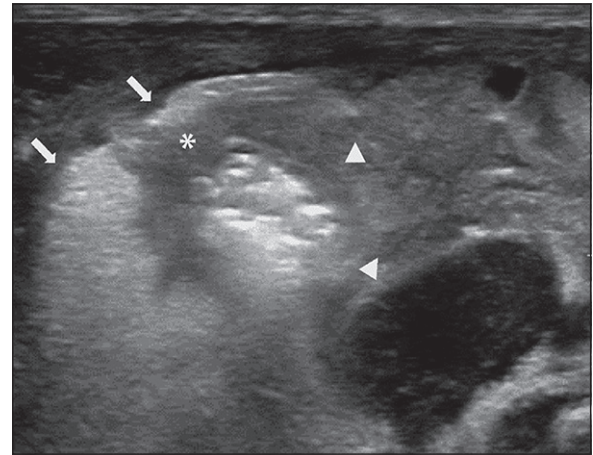


Figure 2b. Abdominal ultrasound scan: it shows thickened retrocecal appendix with apex in a poorly defined hypoechoic area and extraluminal confluent bubbles, suggesting perforation

rated ovary. It was surgically corrected without presenting other complications.

Discussion

Appendicitis in the neonatal period is a rare condition with an estimated incidence of 0.04-0.02%² and its morbidity and mortality are still very high⁶. 75% of the cases occur in men and 52% are preterm newborns⁷.

Several reports have highlighted the clinical similarity with necrotizing enterocolitis, suggesting both a possible association between these conditions and that neonatal appendicitis in the newborn could occur in this setting^{2,5,6,8}. Other associated conditions that have been observed are Hirschsprung's disease, cystic fibrosis, meconium ileus, uni or bilateral inguinal hernia, and cytomegalovirus infection^{3,4}, although, in some reported series, the association with Hirschsprung's disease or cystic fibrosis has not been proved⁹. Due to the low incidence of this condition, the reported series have few cases.

The symptomatology is unspecific. The most common presentation is abdominal distension, which can occur along with sensitivity, irritability, ineffective thermoregulation, food rejection, respiratory distress, and a septic clinical picture^{2,3} with a rapid progression often leading to a bad vital prognosis^{3,4,6,7,10}, reaching in some reports up to 100% of mortality¹¹.

Different factors, both anatomical and environmental, would explain the low incidence in newborns. The appendix is shorter, with a wider funnel-shaped base and a more reclined position. Also contributing are the milk diet without solid food, the lack of coprolith, and the absence of lymphoid hyperplasia^{3,12}.

Among causes, an ischemic environment, and other conditions contributing to this situation are neonatal hypoxia, preeclampsia, umbilical vein catheterization, prematurity, venous insufficiency associated with heart defects, and low blood flow states, which could favor its development¹.

Currently, surgical management is mostly laparoscopic¹⁴ and has both diagnostic and therapeutic objectives. An early diagnosis has a great impact on prognosis, as in septic complications as in survival, considering that due to the clinical features, the diagnosis is usually late^{1,4,5,6,14,15}. To date, some proposals for conservative treatment with antibiotics and without surgery, have no evidence in newborns¹⁶.

The imaging study has significantly contributed to early diagnosis, although until a few years ago, no cases of preoperative use had been reported^{1,7,13}. Recent publications have emphasized the increased use of ultrasound when performed by pediatric radiologists in the diagnosis and complications of appendicitis. However, despite progress, there are few reports on newborns^{17,18}.

In a report of four cases of newborns with preoperative diagnosis of appendicitis by ultrasound, the described findings were ileocecal dilation, bowel wall and cecum thickening, and localized and encapsulated fluid in the right lower quadrant¹⁹. Although ultrasound has limitations, it has proved to be more effective than plain x-rays in evidencing free-flowing intra-abdominal fluid, assessing the bowel wall thickness, and gastrointestinal perfusion¹³. The main ultrasound limitations are related to the method, since being an operator-dependent procedure, its performance is higher in experienced operators and, in cases of late clinical suspicion of the diagnosis, it becomes even more difficult to differentiate this entity from other inflam-

matory processes, such as complicated necrotizing enterocolitis¹⁸. In general, preoperative diagnosis is not frequent⁷. In the reported case, the diagnosis was made before laparoscopic surgical procedure due to these ultrasound findings.

Another challenge in radiological diagnosis is to suspect inflammation of the appendix when it is inside the inguinal canal (Amyand's hernia)²⁰, as well as other complications such as appendicitis concomitant with necrotizing enterocolitis^{5,6,21}, incarcerated hernias where complicated appendicitis underlies¹⁰, or clinical pictures presenting as testicular or ovarian torsion where an inflamed and perforated appendix can be found^{22,23}. The right diagnosis made at the appropriate time allows for a better orientation of surgical strategies.

Although mortality due to appendicitis has decreased between 1901 and 1975 from 78% to 28% between 1985 and 2003, it presents still too high rates due to delayed diagnosis and treatment in a highly vulnerable group as the newborn is²⁴.

Retrospective studies have shown associations of inguinal hernia and appendicitis with inguinal-scrotal signs due to the displacement of the appendix inside the inguinal canal and/or inflammation effects²⁵. In the above case, this form of presentation was not observed since the hernia appeared two weeks later, however, the anatomical condition was present. It could be suggested that when suspecting an inflammatory process in the appendix of newborns, the canals should be carefully examined both with ultrasound and at the time of the laparoscopic exploration.

Conclusion

Although it is very rare, appendicitis should be in-

cluded among the differential diagnoses of acute abdominal disorders in newborns and preterm newborns, considering its high morbidity and mortality rate. Trained pediatric radiologists could improve early diagnosis, allowing for timely management. Inguinal hernia should be included in the conditions to detect, considering the association between neonatal appendicitis and the presence of an inguinal hernia.

Ethical Responsibilities

Human Beings and animals protection: Disclosure the authors state that the procedures were followed according to the Declaration of Helsinki and the World Medical Association regarding human experimentation developed for the medical community.

Data confidentiality: The authors state that they have followed the protocols of their Center and Local regulations on the publication of patient data.

Rights to privacy and informed consent: The authors have obtained the informed consent of the patients and/or subjects referred to in the article. This document is in the possession of the correspondence author.

Conflicts of Interest

Authors declare no conflict of interest regarding the present study.

Financial Disclosure

Authors state that no economic support has been associated with the present study.

References

- Secco IL, Costa T, Moraes ELL, Freire MHS, Danski MTR, Cavalcante RPGV. Neonatal appendicitis: a survival case study. *Rev Bras Enferm*. 2017;70(6):1296-1300. doi: 10.1590/0034-7167-2016-0610.
- Haider F, Ayoub B, Al Kooheji M, Al Juffairi M, Al-Shaikh S. Perforated acute appendicitis with no peritonitis in a premature baby: a case report. *J Med Case Reports*. 2017;11:125. <https://doi.org/10.1186/s13256-017-1289-0>.
- Mammou S, Ayadi I, Hamida E, Marrakchi Z. Acute neonatal appendicitis in a preterm. *African Journal of Paediatric Surgery*. 2015;12(4):294.
- Casal-Beloy I, García-González M, García-Novoa M. Appendicitis, an unusual cause of acute abdomen in neonatal patients. A case report. *Anales del Sistema Sanitario de Navarra*. 2018;41-2.
- Tumen A, Chotai PN, Williams JM, et al. Neonatal Perforated Appendicitis Masquerading as Necrotizing Enterocolitis. *J Neonatal Surg*. 2017;6(2):39. doi: 10.21699/jns.v6i2.482.
- Tumen A, Chotai PN, Williams JM, Myers-Webb A, Krishnan R, Eubanks Iii JW. Neonatal Perforated Appendicitis Attributed to Localized Necrotizing Enterocolitis of the Appendix: A Review. *J Neonatal Surg*. 2017;6(3):60. doi: 10.21699/jns.v6i3.535.
- Romero Rojas N, Sánchez García L. Apéndice aguda neonatal: reporte de un caso. *Anales de la Facultad de Medicina*. 2016;77(4):421.
- Pressman A, Kowar B, Abend M, Steiner Z, Mogilner G. Acute Perforated Neonatal Appendicitis Associated with Chorioamnionitis. *Eur J Pediatr Surg*. 2001;11(3):204-6. doi: 10.1055/s-2001-15481.
- Gregorio-Hernández R, Pérez-Pérez A, Arriaga-Redondo M, Simal I, Sánchez-

- Luna M. Appendicitis abdominal neonatal. Experiencia en un hospital terciario y revisión de la literatura. *Acta Pediatr Esp.* 2017;75(7-8): e138-e140.
10. Fascetti-Leon F, Sherwood W. Neonatal Appendicitis and Incarcerated Inguinal Hernia: Case Report and Review of the Literature. *J Indian Assoc Pediatr Surg.* 2017;22 (4):248-50. doi: 10.4103/jiaps.JIAPS_226_16.
 11. Amin M, Al Jubouri S. Neonatal appendicitis with perforation: a case report. *J Ped Surg Case Reports* 2014;2:353-4.
 12. Marzuillo P, Germani C, Krauss B, Barbi E. Appendicitis in children less than five years old: A challenge for the general practitioner. *World Journal of Clinical Pediatrics.* 2015 4(2):19-24.
 13. Schwartz KL, Gilad E, Sigalet D, Yu W, Wong AL. Neonatal acute appendicitis: a proposed algorithm for timely diagnosis. *J Pediatr Surg.* 2011;46(11):2060-4. doi: 10.1016/j.jpedsurg.2011.07.018.
 14. Stringer MD. Acute appendicitis. *J Paediatr Child Health.* 2017;53(11):1071-6. doi: 10.1111/jpc.13737.
 15. Semerci SY, Kurnaz D, Babayigit A, et al. Neonatal Appendicitis as a Rare Cause of Intestinal Perforation in a Preterm Neonate. *J Coll Physicians Surg Pak.* 2017;27(3):S38-S40. doi: 248.
 16. Yamanaka S, Skarsgard ED, Goldman RD. Conservative therapy for appendicitis in children. *Can Fam Physician.* 2018;64(8):574-6.
 17. Rawolle T, Reismann M, Minderjahn MI, et al. Sonographic differentiation of complicated from uncomplicated appendicitis. *Br J Radiol.* 2019;92(1099):20190102. doi: 10.1259/bjr.20190102.
 18. Reddan T, Corness J, Harden F, Mengersen K. Improving the value of ultrasound in children with suspected appendicitis: a prospective study integrating secondary sonographic signs. *Ultrasonography.* 2019;38(1):67-75. doi: 10.14366/usg.17062.
 19. Si SY, Guo YY, Mu JF, Yan CY. The sonographic features of neonatal appendicitis: A case report. *Medicine (Baltimore).* 2017;96(45):e8170. doi: 10.1097/MD.00000000000008170.
 20. Dhanasekarapandian V, Shanmugam V, Jagannathan M. Amyand's Hernia, Appendicitis, and Undescended Testis: The Amyand's Triad. *J Indian Assoc Pediatr Surg.* 2018;23(3):169-170. doi: 10.4103/jiaps.JIAPS_174_17.
 21. Feldens L, Souza JCK, Fraga JC. Perforation of the cecal appendix in the neonatal period. *J Pediatr (Rio J).* 2018;94(5):567-8. doi: 10.1016/j.jpeds.2018.07.002.
 22. Erginel B, Soysal FG, Celik A, Salman T. Neonatal perforated appendicitis in incarcerated inguinal hernia in the differential diagnosis of testis torsion. *Pediatr Int.* 2017;59(7):831-832. doi: 10.1111/ped.13297.
 23. Perger L, Muensterer OJ. Laparoscopic appendectomy for torsed appendix presenting as an acute abdomen in an infant female. *JSLS.* 2011;15(4):565-7. doi: 10.4293/108680811X13176785204634.
 24. Raveenthiran V. Neonatal Appendicitis (Part 1): A Review of 52 cases with abdominal manifestation. *J Neonat Surg.* 2015;4(1):4.
 25. Raveenthiran V. Neonatal Appendicitis (Part 2): A Review of 24 cases with inguinoscrotal manifestation. *J Neonat Surg.* 2015;4(2):15.