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Michael Bedgood BS, Christine Cortelyou MD, Cynthia Blanco, MD, MSc, Rafael Fonseca, MD, Alvaro Moreira, MD



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## Scrotal Rupture in a Premature Neonate with Cystic Fibrosis as a Consequence of Meconium Periorchitis

Michael Bedgood<sup>1\*</sup> BS, Christine Cortelyou<sup>1</sup> MD, Cynthia Blanco<sup>1</sup>, MD, MSc, Rafael Fonseca<sup>2</sup>, MD, Alvaro Moreira<sup>1</sup>, MD

### Affiliations:

<sup>1</sup>University of Texas Health Science Center (UTHSC), San Antonio, TX; <sup>2</sup>University of Texas Medial Branch (UTMB), Galveston, TX

### Address correspondence to:

Michael Bedgood BS, UTHSCSA- School of Medicine, 7703 Floyd Curl Drive, San Antonio, TX 78229, [bedgood@uthscsa.edu](mailto:bedgood@uthscsa.edu), 469-387-6103 (phone), 210-567-5169 (fax)

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## Abstract

Neonatal meconium periorchitis is a rare condition, with less than 60 cases described in the literature. Of the reported cases, only one describes the complication of a congenital rupture of the scrotum. We present a case of a Hispanic preterm neonate who was diagnosed with cystic fibrosis after scrotal rupture secondary to meconium periorchitis. The neonate was taken to the operating room for exploratory laparotomy and scrotal exploration. No calcification was noted and the patient's left scrotum was surgically packed as well as creating a colostomy. The surgery proved successful and the patient was discharged home on day of life 79. This case of a neonate presenting with meconium periorchitis and scrotal rupture notes the varying degree of initial presentations for cystic fibrosis in a neonate. Successful outcomes for neonates presenting with a ruptured scrotum depend on early clinical assessment.

## Introduction

Neonatal meconium periorchitis (MPO) is a rare condition, with less than 60 cases described in the literature. It is characterized by an in-utero intestinal perforation leading to an extravasation of bowel contents into the scrotum.<sup>1,2</sup> Neonates with MPO typically present with a soft testicular mass or discolored and swollen scrotal area.<sup>3</sup> Of the reported cases, only one describes the complication of a congenital rupture of the scrotum.<sup>4</sup> We present a case of a preterm Hispanic neonate who was diagnosed with cystic fibrosis after scrotal rupture secondary to MPO.

## Case Report

A twin Hispanic male neonate was born at 31 weeks with APGAR scores of 3 and 8 at 1 and 5 minutes, respectively and with a birth weight of 1885 grams. Pregnancy was complicated by polyhydramnios, fetal ascites, poor prenatal care, and the neonate was delivered via Caesarean section for twin-to-twin transfusion syndrome (our patient was the recipient twin). Resuscitation was unremarkable; however, it was noted that the neonate had a grossly distended abdomen and an enlarged left scrotum. A thick, dark discharge, presumed to be meconium, was also found to be exuding from the left scrotum (Figure 1).

In the neonatal intensive care unit, an abdominal radiograph did not demonstrate calcifications, but an ultrasound showed free fluid in the abdomen containing floating debris that

extended into the scrotum. A scrotal ultrasound showed a diffuse echogenic mass within the left scrotal sac.

The neonate was emergently taken to the operating room (OR) for exploratory laparotomy and left scrotal exploration. In the OR, surgeons confirmed an in-utero perforation of the transverse colon with extravasation of meconium through the scrotal sac (Figure 2). The surgeons resected the involved bowel and created a colostomy, while the perforated left scrotum was surgically packed. Daily scrotal packing with ribbon gauze impregnated with Vaseline was performed by the surgical team to facilitate the growth of granulation tissue.

The patient remained on mechanical ventilator support for 14 days and was then transitioned to nasal cannula for 19 days. The neonate returned to the OR on day of life (DOL) 17 for colostomy takedown and he reached full enteral feeds on DOL 20. There were no complications from the surgery, the neonate healed appropriately and was discharged home on DOL 79. DNA testing established diagnosis of cystic fibrosis in both neonates (homozygous F 508 mutation). The neonate had follow up at 4 months in clinic with minimal scarring and no drainage.

### **Discussion / Review of Literature**

Meconium periorchitis (MPO) is an unusual complication of meconium peritonitis. Since the discovery of meconium peritonitis in 1953, the phenomenon of MPO has been described in fewer than 60 cases in the literature.<sup>5,6</sup> Our case is exceptionally rare as there has only been one documented patient over the past 2 decades who presented with extravasation of bowel contents through the scrotum.<sup>4</sup> There have only been three reported cases of MPO in patients with cystic fibrosis and none of these cases have presented with congenital scrotal rupture.<sup>5,6,7,8</sup>

The clinical presentation of MPO is variable, but typically it presents in-utero or at birth with scrotal swelling due to the presence of meconium. The displaced meconium initiates an inflammatory reaction that may begin within 24 hours and results in fibrosis and calcifications.<sup>6</sup> Forms of peritoneal calcifications include localized and generalized fibroadhesive type. Localized fibroadhesive type is the most common form, resulting in linear peritoneal calcification, while the generalized fibroadhesive type is associated with ascites.<sup>9</sup> Scrotal calcifications, with or without these peritoneal calcifications, are pathognomonic findings on perinatal ultrasonography.<sup>8</sup>

If MPO is not clinically apparent at birth, it will present in a milder form later in the neonatal period often resembling hydroceles, which calcify with time. These patients have been noted to return to their primary care provider within the first two years of life with a palpable scrotal mass.<sup>9</sup> This delayed presentation allows time for the meconium in the scrotal sac to calcify and become adhered to the testicle forming a hard, nodular, solid mass. Previous case reports demonstrate that this clinical finding makes it difficult to determine if the mass is intra- or extra- testicular often leading to surgical exploration.<sup>6</sup> Table 1 shows a comparison of meconium periorchitis cases, their presentation, management of the scrotal mass, and their outcome. While most cases are discovered during prenatal ultrasound, some, including our case, are detected at birth or after. Most of the cases resulted in surgical removal of the mass, and only two patients were found to have cystic fibrosis.

The successful outcome of our patient was due to several factors including: early assessment of clinical presentation, no testicular involvement, and no additional surgery required for repair of the ruptured scrotum. At the time of the surgery an ultrasound was performed showing bilateral descended testicles, normal epididymis and good perfusion indicating a lack of testicular involvement. Testicular involvement can lead to complications not immediately measurable including testicular atrophy, infertility, or torsion. Additional surgery could have resulted in further complications including infection, adhesions or excessive bleeding. With scrotal rupture present, the bowel perforation was able to be identified and repaired while sparing testicular surgery. Previous literature has noted that finding a source for the scrotal mass can lessen confusion for a scrotal tumor leading to unnecessary orchiectomy.<sup>10</sup>

## **Conclusion**

When meconium periorchitis with congenital rupture of the scrotum is found, it is important to consider cystic fibrosis as a cause. These findings are relevant to all neonates that present with atypical features of cystic fibrosis such as scrotal rupture in order to expedite their diagnosis and ensure proper follow up care. This identification may spare the need for unnecessary orchiectomy and lead to more successful outcomes.

**Bulleted Learning Points**

- Meconium periorchitis with congenital rupture of the scrotum can be the initial presentation of cystic fibrosis in neonates.
- Meconium periorchitis is an in-utero intestinal perforation leading to extravasation of bowel contents into the scrotal sac.
- Recognizing congenital rupture of the scrotum can help identify bowel perforation that may be treated surgically based on patient factors with colostomy and extravasation of the meconium through the scrotal sac.

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**Figure 1.** Initial appearance of patient's scrotum



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**Figure 2.** Intrauterine perforation of transverse colon



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**Table 1.** Comparison of case presentations of meconium periorchitis

Year	Author	Age	Presentation	Management	Outcome
2016	Bedgood, et al	31 weeks gestation	Scrotal rupture	Surgery at presentation	Cystic Fibrosis
2013	Alanbuki, et al <sup>10</sup>	2 months post term	Scrotal mass	Surgery at presentation	Uneventful
2009	Regev, et al <sup>3</sup>	39 weeks gestation	Hydrocele	Surgery at 5 months post term	Uneventful
2009	Regev, et al <sup>3</sup>	36 weeks gestation	Hydrocele	Conservative	Uneventful
2003	Sofeman, et al <sup>7</sup>	37 weeks gestation	Hydrocele	Conservative	Cystic Fibrosis