

## CASE REPORT

# A rare case of necrotizing fasciitis after early infant male circumcision

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## Abstract

Even though circumcision is low-risk, complications may occur, necessitating careful management. Here, we describe a 5-month-and-28-day-old infant who developed necrotizing fasciitis following Plastibell circumcision. This report emphasizes that clinicians should use standard equipment and appropriate-sized rings to prevent these unusual complications following circumcision.

## KEYWORDS

anti-bacterial agents, circumcision, infant, necrotizing fasciitis

## 1 | INTRODUCTION

Male circumcision is a minor surgical removal of the skin surrounding the glans penis.<sup>1</sup> Circumcision could be conducted using various methods, including the Gomco clamp, dorsal slit, and Plastibell. The Plastibell circumcision device is a transparent plastic ring with varying diameters and a handle designed for male circumcision. The ring, which comes in various sizes, has a circular groove with a deep ridge.<sup>2</sup> As one of the most frequent elective surgeries in medicine, circumcision is carried out for several reasons, including treating an underlying disease or as indicated by cultural beliefs.<sup>1</sup> Only a minimal risk of complications is associated with this procedure. Infection, bleeding, injury to the glans, staphylococcal scalded skin syndrome (SSSS), and necrotizing fasciitis are among the complications.<sup>2,3</sup> Necrotizing fasciitis is a severe condition in which the subcutaneous layer and superficial fascia are infected, causing

erythema, swelling, and pain in the affected area. Despite this being a relatively rare condition in newborns, it requires early diagnosis, appropriate attention, and immediate treatment once it is discovered. We report the case of a 5-month-and-28-day-old infant who developed necrotizing fasciitis (NF) after Plastibell circumcision.

## 2 | CASE PRESENTATION

### 2.1 | History

Our patient was a boy of 5 months and 28 days of age and 7.2 kg at the time of admission. He was born by natural delivery (NVD) at 37 weeks.

Three days before admission, he had undergone ring circumcision. Two days later, his mother discovered him unwell, developing erythema, swelling in his genital area,

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and purulent discharge on his diaper; further, they realized that the infant's scrotum had changed color to black, purple. Currently, the infant refuses to eat and gradually develops oral feeding intolerance.

Additionally, his body temperature increased during these days to 38°C. Due to the infant's agitation at home, his parents administered opioids to relieve him of his symptoms. Afterward, a general practitioner visited the infant and incorrectly prescribed betamethasone ointment. On April 20, the infant was taken to our hospital.

## 2.2 | Physical examination

His condition on arrival was septic; he had a body temperature of 38°C, a systolic blood pressure of 80 mm of mercury, and a heart rate of 140. The scrotum was swollen and inflamed on physical examination, as shown in (Figure 1).

The inflammation had spread to the adjacent tissues and the perineal region over the pubis symphysis. Moreover, excessive purulent discharge was noted on superficial palpation of the penis. Also, examining the scrotal skin revealed erythema and heat; a closer checking of the scrotum revealed necrotic tissue. On palpation of the scrotum, crepitation was detected.

## 2.3 | Laboratory data

In the laboratory, he had a WBC count of 24,700/mm<sup>3</sup> (normal range: 9100–30,400/mm<sup>3</sup>) (PMN: 50.4%), a hemoglobin level of 13.9 g/dl (normal range: 10–13 g/dl), and a platelet level of 490,000/mm<sup>3</sup> (300,000–700,000/mm<sup>3</sup>). He had a CRP of 34 mg/dl (normal range: <0.08 mg/dl) and an ESR of 75 mm/h. In addition, his VBG revealed a pH of 7.41 (normal range: 7.35–7.45), a pCO<sub>2</sub> of 21.6 (35–45 mmHg), and an HCO<sub>3</sub> of 13.7 (normal range: 20–28 mEq/L). Furthermore, his blood culture revealed no growth. However, his skin smear and culture were positive for gram-positive cocci. His laboratory data showed



FIGURE 1 Three days after plastibell circumcision.

improvement during the hospital stay with a WBC of 12,900/mm<sup>3</sup> (PMN:40%), CRP of 25 on Day 5, WBC of 4700/mm<sup>3</sup> (PMN: 20%), CRP of 8, and ESR of 75. Table 1 shows an overview of the laboratory data during the hospital stay.

## 2.4 | Differential diagnosis

Toxic shock syndrome.

Orchitis.

Testicular gangrene following torsion.

Cellulitis.

## 2.5 | Treatment/Plan

Regarding the circumcision procedure, we did not have a detailed report since the operation was conducted in a private center, and we did not have access to the medical record. However, it was evident that the ring size was not appropriate for age. Given the complications, it is probable that the operation was not conducted in a sterile condition and with proper technique. Having observed that the infant had been given opioids 3 days ago, we had suspicions regarding sufficient urinary output. For this purpose, we attempted to insert a Folley catheter at the emergency department. On the first attempt, the ring was separated. As a result of the patient's urgent condition, we initiated ampicillin-sulbactam and vancomycin, and he was immediately taken to the operating room. The scrotum was split open during the operation, revealing two swollen testicles and necrotic tissue adjacent to the testicles and extending above the inguinal region. Considering the presence of edema as an indicator of the need for surgical debridement, the necrotic tissues were debrided in several debridements.

Further, we used normal saline and acetic acid to enhance the surgical debridement. The dissection did not invade the tunica vaginalis (Figure 2). Following this, wound irrigation and packing were performed daily. Due to peripheral edema, the delayed primary closure was postponed until the peripheral edema subsided (Figure 3).

## 2.6 | Outcome

The infant was discharged after fifteen days of hospitalization. On discharge, he was afebrile and in good general condition. Furthermore, the extent of edema decreased after surgical and medical treatment. In the laboratory, there was no evidence of active inflammation (i.e., elevated CPR).

**TABLE 1** Overview of the laboratory data changes during the hospital stay

	WBC (mm <sup>3</sup> )	PMN	CRP	ESR
Admission	24,700	51%	34	75
Day 5	12,900	40%	25	–
Discharge	4700	20%	8	–

**FIGURE 2** Ten days after circumcision.

### 3 | DISCUSSION

Circumcision is currently done using a variety of techniques. The primary methods include the dorsal slit, Gomco clamp, Mogen clamp, bone cutter, and Plastibell. Circumcision performed using ring tools is a popular and commonly selected approach due to its many advantages, including its speed, simplicity, less traumatic technique, less blood loss, decreased complication rates, and high cosmetic satisfaction. Plastibell ring devices are used on children, while PrePex and Shang Ring are used on people older than 18.<sup>4</sup> On the contrary, ring devices used for circumcision have been shown to have a minimal advantage in terms of postoperative complications over other methods; their most significant advantages are the short operation duration, high family satisfaction in terms of cosmetic appearance, and the ability to be quickly learned and performed by assisting health personnel in countries lacking sufficient professional health personnel.<sup>5</sup> However, it is a must to be familiar with open circumcision techniques and have expertise doing this procedure, especially in

**FIGURE 3** Ten days after final repair.

bleeding difficulties and the absence of equipment of the correct size.

Necrotizing fasciitis is a sporadic condition in adults and even rarer in children, with a prevalence of 0.08 per 100,000. However, studies have shown that the incidence of this disorder is on the rise among children.<sup>4,6</sup> The disease could be developed in the presence (secondary) or the absence (primary) of underlying conditions. Most children who develop necrotizing fasciitis are otherwise healthy, unlike adults. Among the conditions that may result in necrotizing fasciitis are trauma, immunosuppression, infections such as balanitis, and, rarely, circumcision.<sup>5,7</sup> In our case, the patient developed necrotizing fasciitis two days after circumcision. We believe that the very surgical techniques (i.e., performing surgery in non-standard conditions and using rings of the wrong size) have contributed to necrotizing fasciitis. Moreover, the incorrect prescription of betamethasone has further deteriorated the states, and the opioid administration by his family has falsely subsided his symptoms.

Necrotizing fasciitis begins with a localized infection of soft tissues. The bacteria then spread to the fascia that separates the muscles. After this, the accumulated bacteria, toxins, and cytokines enter the bloodstream, resulting in systemic toxicity, thrombus formation, ischemia, and necrosis.<sup>7</sup> Because the responsible bacteria mainly exhibit their effects in the subcutaneous layer, the skin is expected to be spared in the early stages of the disease. Nevertheless, these findings are not encouraging; studies indicate that necrosis advances at a rate of 1 inch/hour in the tissues beneath. Therefore, it should always be assumed that the

affected area is more significant than observed on physical examination.<sup>8</sup> Typically, necrotizing fasciitis is recognized with severe pain compared to the physical examination, skin numbness, and symptoms of sepsis, that is, tachycardia, hypotension, and altered mental status. In the early stages of necrotizing fasciitis, there may be tenderness and erythema, making a misdiagnosis of cellulitis more likely. Despite some studies describing “severe pain out of proportion” as the most common finding in children,<sup>9</sup> a systematic review reported tenderness as the most common finding in necrotizing fasciitis in adults and children. Concerning the site of involvement, the etiology plays a central role.<sup>10</sup>

Furthermore, necrotizing fasciitis in adults has generally been reported to affect the extremities and the perineum, whereas, in children, there is no consensus as to the most common site, as some studies have focused on the extremities,<sup>10</sup> others on the lumbar region,<sup>5</sup> and yet others on the lateral abdominal wall.<sup>9</sup> In our case, the involved area was the perineum. It is thought that there are two types of necrotizing fasciitis: Type I, which is caused by a mix of gram-positive, gram-negative, and anaerobic bacteria; and Type II, which is caused by a monobacterial agent, most commonly group A beta-hemolytic streptococcal bacteria. Type I usually occurs in the presence of underlying conditions, while Type II occurs in healthy individuals.<sup>11</sup> As our patient was in an emergency, we had to initiate therapy before the cultures could be prepared. Given the patient's lack of underlying disease, we started 1 million U of penicillin IV every four hours. The results of culture later revealed group A beta-hemolytic streptococcal bacteria.

Necrotizing fasciitis is commonly diagnosed clinically. Radiology may also be helpful, with MRI as the best modality to detect fluid collection in soft tissue; however, due to the availability of ultrasonography at the bedside, this method may be a procedure if an expert performs its better option for pediatrics. Nevertheless, a prompt diagnosis is only possible through surgical exploration, leaving the analysis mainly up to the clinician's judgment. In light of the grave concerns regarding this condition, efforts have been made to establish criteria for diagnosing necrotizing fasciitis; however, efforts to develop scoring systems for pediatric patients are minimal.<sup>12</sup> Extensive surgical debridement of the necrotic tissues and fascia is the main component of treatment. The timing of surgery should be highlighted as the sole factor influencing the result.<sup>5</sup> In addition, patients should receive aggressive fluid resuscitation, antibiotic treatment, and pain management. On admission, the patient underwent surgical removal of necrotic tissues and continued to receive treatment under close observation over the following sixteen days.

The patient was ultimately discharged in good health (Figure 2).

## 4 | CONCLUSION

Pediatric necrotizing fasciitis is a rare but often fatal bacterial infection of the subcutaneous tissues and the superficial fascia. Clinically, it is characterized by tenderness, erythema, and pain out of proportion. The most important aspects of neonatal NF care are high suspicion for diagnosis, prompt aggressive surgical intervention, adequate antibiotic treatment, and supportive care. It is infrequent for this condition to develop after circumcision, so it is advisable to perform the procedure in a standard facility and use rings of an appropriate size. Furthermore, parents of children who have undergone Plastibell circumcision should be aware of the risk of necrotizing fasciitis and encouraged to visit an emergency room in case complications arise.

## AUTHOR CONTRIBUTIONS

Fariba Zabihi involved in manuscript preparation, editing, and review. Saeid Jamalie Bastami involved in data interpretation and conduct. Khashayar Atqiaee involved in reporting and planning.

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## CONFLICT OF INTEREST

The authors declare no competing interests relating to this original work.

## DATA AVAILABILITY STATEMENT

The datasets are available from the corresponding author on reasonable request.

## ETHICAL APPROVAL

We confirm that all named authors have read and approved the manuscript. The protection of the intellectual property associated with this manuscript has been our consideration.

## CONSENT

The authors confirmed that they had gotten all proper patient written consent formats. The parents have given their consent for their images and other clinical information to be reported in the form. The parents were informed



about the confidentiality of the names and initials, and efforts would be made to hide their identity.

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