# CASO CLÍNICO / CASE REPORT

# Celulite Inguinal Como Manifestação de Infeção Tardia por Streptococcus do Grupo B

Inguinal Cellulitis as Manifestation of Late-Onset Group B Streptococcal Infection

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## Resumo

O *Streptococcus* do grupo B é considerado um dos principais agentes de infeção perinatal, tanto precoce como tardia, e pode ser responsável por um amplo espetro de doença sistémica ou localizada. Apresenta-se o caso clínico de uma lactente com 4 semanas de idade, previamente saudável, com celulite de localização inguinal associada a bacteriemia por *Streptococcus* do grupo B e discutem-se a apresentação clínica, as alterações nos exames complementares de diagnóstico e a evolução desta entidade clínica, com base numa revisão da literatura. O caso apresentado destaca a celulite como uma forma de apresentação rara de infeção tardia por *Streptococcus* do grupo B, demonstrando que pode ser o único marcador de doença invasiva em crianças saudáveis.

**Palavras-chave:** Canal Inguinal; Celulite; Infeções Estreptocócicas; Lactente; Streptococcus agalactiae; Transtornos de Início Tardio

#### **Abstract**

Group B streptococcus is one of the leading pathogens of both early- and late-onset neonatal infection, and can cause a wide spectrum of focal and non-focal disease. The authors report the case of a previously healthy four-week-old baby with inguinal cellulitis associated with group B streptococcus bacteraemia, and discuss the clinical features, laboratory findings, and outcome of this clinical entity based on a review of the literature. This case highlights cellulitis as a rare presentation of late-onset group B streptococcal infection, demonstrating that it may be the only marker of invasive disease in healthy children.

**Keywords:** Cellulitis; Infant; Inguinal Canal; Late Onset Disorders; Streptococcal Infections; Streptococcus agalactiae

### Introduction

Group B streptococcus, or Streptococcus agalactiae, is an encapsulated Gram-positive diplococcus that produces a narrow zone of beta-hemolysis on blood agar and is usually resistant to bacitracin. The organism began to attract attention in the 1970s as a cause of maternal and neonatal disease.<sup>1-3</sup>

In newborns and infants, group B streptococcal infection classically presents as early-onset disease (EOD, before

seven days of age), late-onset disease (LOD, up to 89 days after birth, but generally within one month), and late, late-onset disease (LLOD, usually in the context of preterm birth, up to six months of age).<sup>1-5</sup> EOD is typically related to the presence of group B streptococcus in the maternal genital tract, which can infect the amniotic fluid and the neonate during delivery. By contrast, LOD and LLOD are thought to be acquired from maternal, nosocomial or community sources.<sup>1,5,6</sup>

As group B streptococcal infection often presents with nonspecific clinical signs and is associated with underlying bacteraemia, prompt investigation and treatment are required.<sup>1</sup>

# **Case Report**

A four-week-old female infant with fever, poor feeding and irritability lasting for six hours was brought to the emergency department. There were no respiratory symptoms. The gestation was monitored without complications. Maternal serology tests, including hepatitis B virus surface antigen (HBsAg), human immunodeficiency virus (HIV), cytomegalovirus, rubella virus and toxoplasmosis in the third trimester showed no evidence of acute infection. Screening of maternal group B streptococcus colonisation performed at 35 weeks of gestation was negative. Cultures of the mother's urine during pregnancy were also negative. The baby was born at 40 weeks by caesarean section. Membranes ruptured <12

hours before delivery. The immediate neonatal period was uneventful. She was exclusively breast-fed since birth and was thriving well until this event.

On examination, the infant was irritable and with an axillary temperature of 39°C. Her vital signs were within normal ranges. Significant findings included erythema, with marked swelling, warmth and induration of the left inguinal region for several hours (Fig. 1). She had pain and decreased motion of the left lower limb.

The complete blood count revealed 7600 white blood cells/ $\mu$ L with 70.6% neutrophils and 11.7% lymphocytes, haemoglobin 10.9 g/dL and platelet count of 242 000/  $\mu$ L. Serum C-reactive protein concentration was increased (22.4 mg/L). There were no white blood cells or nitrites detected by urine analysis.

An ultrasound examination of the left inguinal area showed an increase in the thickness and echogenicity of the subcutaneous tissue, associated with the presence of small hypoechoic laminae indicating accumulation of fluid, with a maximum diameter of 14 mm and a maximum thickness of 2 mm. There were also millimetre-sized inguinal lymph nodes.

Lumbar puncture was tried without success. A diagnosis of cellulitis was established and she was hospitalised and treated empirically with intravenous ampicillin and cefotaxime.

Within 24 hours the infant became afebrile and less irritable. Feedings improved progressively. Clinical regression of the cellulitis was noted three days after the start of antibiotics. Group B streptococcus was isolated in the blood culture.

Ampicillin and cefotaxime treatment was maintained for 14 days. The source of infection remained unidentified. Follow-up visits have been conducted since discharge. The hearing screen performed at six months of age was normal. One year later the infant has normal neurodevelopment.



Figure 1. Inguinal cellulitis.

## **Discussion**

Cellulitis is a rare presentation and most often associated with late-onset group B streptococcal infection. <sup>1,6</sup> The peak incidence generally occurs at an average of five weeks of age. <sup>7</sup> In older children, cellulitis is most commonly a discrete, local infection caused by group A beta-haemolytic streptococci or Staphylococcus aureus. In infants less than 3 months of age, cellulitis is most commonly caused by group B streptococcus and is associated with invasive disease in a significant number of cases. <sup>7</sup>

The most commonly described sites of cellulitis are the face and scalp, mainly in the preauricular, submandibular or submental areas.<sup>6,7</sup> Cellulitis localized in the inguinal region as occurred in this case is rarely reported.<sup>8</sup> Although the spectrum of presentation is wide, there are some common features described, such as poor feeding, irritability, fever, and localized swelling of the skin,<sup>6</sup> as occurred in the present case.

In infants younger than three months, cellulitis has been associated with bacteraemia in up to 90% and meningitis in about 25% of cases.

The origin of bloodstream infection in LOD is not clearly elucidated.7 One hypothesis postulates that group B streptococcal disease is associated with ingestion of infected breast milk.<sup>1,4,6-9</sup> According to the literature, the presence of group B streptococcus in the maternal genital tract at birth is the major determinant of colonisation and infection in the infant.<sup>2</sup> It was recently reported that up to 64% of mothers with babies diagnosed with LOD carried group B streptococcus at the rectovaginal site, suggesting the importance of vertical transmission of group B streptococcus from the colonised mother to the patient.<sup>1,5</sup> It is now standard practice in Portugal and many other countries for all pregnant women to be routinely tested for group B streptococcus at 35 to 37 weeks of gestation, unless their urine cultures were already positive in the current pregnancy. Although this test was performed at the recommended period in the reported case, group B streptococcal colonisation during pregnancy can be intermittent, so maternal culture status can change after screening.3 In addition, the clinical utility of prenatal culture decreases when it is performed more than five weeks before delivery. Furthermore, there is strong evidence that universal screening combined with intrapartum antibiotic prophylaxis is an important measure to prevent early-onset group B streptococcal disease. However, it does not prevent late-onset disease. 2,3,5,9,10 Horizontal transmission from community or nosocomial sources is another hypothesis for infection, although much less common.2

LOD incidence in Europe ranges from 0.10 to 0.24 per 1000 live births (an incidence 1.5-4 times lower than that associated with EOD), and case fatality rates range from 2% to 8%.5

Unlike EOD, in LOD there are usually no maternal obstetric or nursing complications.

It is difficult to determine clinically which neonates have central nervous system disease, because the clinical presentation can be the same as that of bacteraemia or pneumonia. Only examination of the cerebrospinal fluid can exclude the diagnosis.<sup>2</sup> These infants are generally mildly ill on presentation but can progress rapidly to shock if not treated promptly.

The absence of seizures, poor perfusion, neutropaenia and hypotension can be predictive of a better outcome, as observed in the case reported.<sup>2</sup> A definitive diagnosis of invasive group B streptococcal infection is based on isolation of the organism from a normally sterile body site. C-reactive protein level and white blood cell count are not specific to a diagnosis of group B streptococcus, but may be helpful in determining the extent of the disease and the clinical response to treatment.

In general, group B streptococcus is susceptible to ampicillin, penicillin, meropenem, imipenem, vancomycin, cephalosporins, and levofloxacin.<sup>2</sup> As it was not possible to exclude meningitis in this case, the treatment was maintained for 14 days.

Infection may recur, although this is not common.<sup>2,10</sup> Appropriate antibiotic therapy does not always eliminate mucosal colonisation; the infant can be reexposed in the community and invasive infection does not result in protective levels of antibodies to capsular polysaccharides.<sup>2</sup> Although LOD has been reported to be common among infants with underlying conditions such as premature birth, immunocompromised status or trauma, or using medical devices, none of these underlying medical condition predisposed this infant to invasive group B streptococcal infection. Recent reports including the present case underscore the risk of group B streptococcal infection among previously healthy infants beyond the neonatal period.<sup>1,5</sup> Group B streptococcus should be considered as a possible aetiological agent in young infants presenting nonspecific signs of systemic infection and localized cellulitis. Inguinal cellulitis is an uncommon signal of group B streptococcal disease and may be a clear indicator of underlying bacteraemia.

This case highlights that healthy children under three months of age can also be affected with invasive group B streptococcal disease and that universal culture-based screening of all pregnant women between 35 and 37 weeks gestation has no measurable impact on LOD presenting as bacteraemia and cellulitis. Thus, it is important for clinicians to be aware of unusual manifestations of late-onset group B streptococcal invasive infection, which requires a different diagnostic course and treatment.

#### WHAT THIS CASE TEACHES

- Cellulitis in newborn and infants is a rare presentation and is most often associated with late-onset group B streptococcal infection.
- Previously healthy children can also be affected with invasive group B streptococcal disease beyond the neonatal period.
- As group B streptococcal infection often presents with nonspecific clinical signs and is associated with underlying bacteraemia, it is important to keep group B streptococcus as part of the differential diagnosis when evaluating infants who show late signs consistent with infection.

#### **Conflitos de Interesse**

Os autores declaram a inexistência de conflitos de interesse na realização do presente trabalho.

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## Proteção de Pessoas e Animais

Os autores declaram que os procedimentos seguidos estavam de acordo com os regulamentos estabelecidos pelos responsáveis da Comissão de Investigação Clínica e Ética e de acordo com a Declaração de Helsínguia da Associação Médica Mundial.

## **Confidencialidade dos Dados**

Os autores declaram ter seguido os protocolos do seu centro de trabalho acerca da publicação dos dados de doentes.

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