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CASE REPORT

A common diagnosis in an uncommon age: acute neonatal parotitis case report

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Abstract

Acute neonatal parotitis is a rare disease, usually with a clinical diagnosis. Parotid swelling and other inflammatory signs are essential characteristics. Purulent discharge of Stensen's duct is pathognomonic of suppurative parotitis. *Staphylococcus* aureus is the most common pathogen, although other organisms may also be involved. We describe the case of a 10-day-old term newborn who presented with subtle preauricular swelling and erythema. In this case, the ultrasound was essential since obvious inflammatory signs and purulent drainage only occurred later, and there were no known risk factors. No microorganism was isolated since it was not feasible to collect an uncontaminated sample. Empiric antimicrobial therapy was initiated, with consequent clinical improvement. This condition should be considered in the differential diagnosis of parotid region swelling. Early suspicion, diagnosis and appropriate combined antibiotics are essential for a rapid and complicationsfree recovery.

Keywords: Acute parotitis. Case report. Newborn. Suppuration. Ultrasound.

Um diagnóstico comum em uma idade incomum: relato de caso de parotidite neonatal aguda

Resumo

A parotidite aguda neonatal é uma doença rara, habitualmente com diagnóstico clínico. O edema da parótida e outros sinais inflamatórios são características essenciais. A drenagem purulenta pelo ducto de Stensen é patognomónico da parotidite supurativa. Os autores descrevem um caso de um recémnascido com 10 dias de vida que se apresentou com discreto edema e eritema da região pré-auricular. Neste caso, a ecografia foi essencial para o diagnóstico, uma vez que os sinais inflamatórios mais exuberantes e a drenagem purulenta só ocorreram mais tarde e não existiam fatores de risco conhecidos. Não foi isolado nenhum microrganismo uma vez que não possível colher uma amostra não contaminada. Foi iniciada antibioterapia empírica com consequente melhoria clínica. Esta patologia deve integrar o diagnóstico diferencial de edema da região parotídea. A suspeita, o diagnóstico e o tratamento precoces, com antibioterapia combinada, são essenciais para uma recuperação rápida sem complicações.

Palavras chave: Parotidite aguda. Caso clínico. Recém-nascido. Supuração. Ecografia.

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Keypoints

What is known

– Acute parotitis is a rare disease in newborns.
– Swelling of parotid gland is an universal sign and discharge from the Stensen's duct is ered in the parotitis.
– Surgical drainage is rarely necessary. Early antibisubtle clinical findings

What is added

– ANP can occur in a newborns without risk factors.
– Acute neonatal parotitis should always be considered purulent differential diagnosis of parotid region pathognomonic of suppurative swelling.
– Ultrasound can be fundamental, particularly otics are fundamental for a good outcome and to avoid complications.

Introduction

Although salivary gland infections are uncommon in the neonatal period, the parotid is the most commonly involved gland[1,2]. Acute neonatal parotitis (ANP) is a rare disease, first described in 1878[1]. Since then, approximately 150 cases have been reported, nine of them in Portugal[3,4]. Prematurity and dehydration are considered major risk factors[5]. Parotid swelling is a primary sign of ANP and purulent discharge from the Stensen's duct is pathognomonic of suppurative parotitis[6,7]. *Staphylococcus aureus* is the most common agent and systemic antibiotic treatment is the first-line therapy[7]. The prognosis is favourable with rare recurrence[8].

The authors describe a case of acute neonatal parotitis in a 10-day-old girl and present a brief review of this pathological condition.

Case report

A 10-day-old breastfed female newborn was admitted to hospital with subtle right preauricular swelling, slight redness and pain described as a "cry-on-touch", with < 12h of evolution. She was born at full-term by induced vaginal delivery due to maternal cholestasis, after an uneventful pregnancy with negative serologies (including for Human Immunodeficiency Virus (HIV)) and normal ultrasounds. Maternal vaginal and rectal group *B Streptococcus* cultures were negative. During the three-day hospital stay, she presented neonatal jaundice, which required 24h phototherapy. There was no history of trauma to the infant's face or head, mother's mastitis, or other maternal skin infection. The weight progression was appropriate with exclusive breastfeeding.

On physical examination, she was afebrile (36.7ºC), clinically well, hydrated, active and reactive. A slight facial asymmetry with enlargement of the right peri-auricular area and effacement of the mandibular angle was noted, with apparent discomfort on palpation (Fig. 1). It was not possible to individualize any mass or ganglion. The rest of the physical exam was unremarkable, including the oral cavity.

A local ultrasound scan was requested, revealing an enlarged right parotid gland with heterogeneous texture and increased vascularity, subcutaneous tissue thickening and internal and adjacent reactive lymph nodes (Fig. 2). No collections were observed. These findings were consistent with acute parotitis. Laboratory tests showed a hemoglobin of 16.6 g/dL, white blood cell count of 18.6x109/L (with 53.4% neutrophils, 26.8% lymphocytes and 16.3% monocytes), C-reactive protein 2.8 mg/L (normal) and procalcitonin 0.15 μg/L (normal). A blood culture was obtained.

The newborn was hospitalized and started intravenous empirical antibiotic therapy with flucloxacillin (150 mg/Kg/day) plus gentamicin (4 mg/Kg/day). In the following hours, the general state worsened with intermittent whimpering and a slight elevation of axillary temperature (37.7ºC). The facial asymmetry became more notorious with exacerbation of erythema, firm swelling and warmth (Fig. 3).

Pus from Stensen's duct came out after applying pressure to the parotid gland. Unfortunately, it was not possible to collect an uncontaminated sample. She remained afebrile for the rest of her hospital stay, and after two days of treatment, the local inflammatory signs began to improve.

On the fourth day of hospitalization, she repeated laboratory tests, with negative C-reactive protein and procalcitonin, serum amylase concentration of 7 U/L (normal) and gentamicin value within the therapeutic range (0.2 mg/L). The blood culture was negative. She completed six days of intravenous antibiotic therapy and initiated oral amoxicillin-clavulanate (80 mg/Kg/day). She was discharged home on the 7th day with slight facial asymmetry and maintained oral therapy for an additional seven days. During hospitalization and afterwards, breastfeeding was never interrupted.

At the follow-up examination, 20 days later, she was in good health and her parotid swelling had resolved. A revaluation ultrasound was performed, showing symmetrical parotid glands with no abnormalities. Five months after diagnosis, there is no record of complications or recurrence.

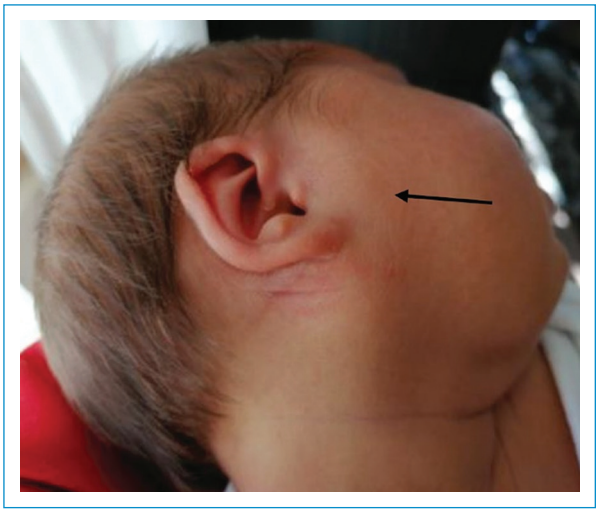

Figure 1. Subtle swelling (arrow) in the right periauricular region on examination.

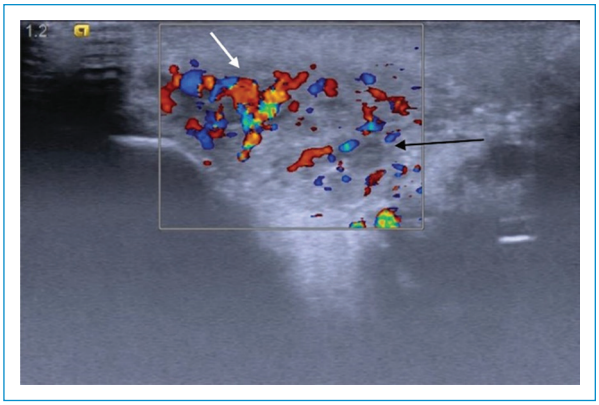

Figure 2. Ultrasound showing an enlarged right parotid gland with heterogeneous texture (black arrow) and increased vascularity (white arrow).

Discussion

Acute neonatal parotitis is an unusual disease with a prevalence of 3.8-14/10000 admissions in different studies[4,7,9]. It was considered a vanishing disease, however, sporadic cases have been reported[4].

Its incidence is three times higher in males, predominantly unilateral (each parotid gland is equally involved)[5,10] and occurs between the 7th and 14th days[3,6] until four weeks of age[9]. Prematurity (associated with prolonged hospitalization and trauma of the oral cavity by tube feeding), dehydration, ductal obstruction and structural abnormalities are all known risk factors[5,9-11]. These factors may cause stasis or reduce salivary secretion, contributing to the development of parotitis[12]. Maternal breast abscess, maternal treatment with methyldopa[3,7], transmission of bacteria through infected breast milk or contaminated formula have also been associated with parotid gland infection[3,9]. In our case, the patient was a full-term female newborn and no risk factors were identified.

The most common route of parotid gland infection is by retrograde bacterial flow from the oral cavity into the gland via Stensten's duct; however in some cases, parotitis occurs via hematogenous in the con text of sepsis[7,13]. In our case, the presence of purulent discharge and the absence of bacterial isolation in the blood culture supports the diagnosis of suppurative parotitis via retrograde route of the oral cavity.

S. aureus is the most frequently isolated pathogen (55% of cases). Other less common agents are other Gram-positive cocci (*Viridans streptococci, Streptococcus pyogenes, Peptostreptococcus and coagulase-negative Staphylococcus)* (22%), Gramnega tive bacilli (*Escherichia coli, Klebsiella pneumoniae* and *Pseudomonas aeruginosa)* (16%) and rarely anaerobic bacteria, such as *Bacteroides melaninogenicus* and *Fusobacterium nucleatum*[5,7,10,14].

Neonatal parotitis is a clinical diagnosis, with swelling of parotid gland being an universal sign and local inflammatory signs (erythema and tenderness) occurring less frequently[7,10]. Fever (found in less than half of the cases), food refusal, irritability and other non-specific symptoms may also occur[15]. Purulent drainage from the Stensen's duct is pathognomonic of suppurative ANP[6,7], despite it might not be noted at presentation, as was the case in this newborn.

Laboratory tests are generally non-specific. A leukocyte count above 15 × 10^9/L with neutrophil predominance is the most frequent finding (71% of patients)[4,10]. Elevated serum amylase was detected only in 25% of cases, which may be related to the immaturity of salivary isoenzyme activity in infants[5,7]. Cultures of blood and purulent material from Stensen's duct are useful for diagnosis and therapy guidance[15]. Breast milk culture should be performed when there is a history of mastitis or maternal breast abscess, as it may be the source of the infection[3,13].

Ultrasound is the most used imaging technique to confirm the diagnosis. Similar to what was found in the reported case, it often reveals an enlarged heterogeneous gland, with hypoechoic areas and increased vascularity[6,7,16,17]. Additionally, ultrasound and other imaging techniques are particularly relevant to differential diagnosis, in cases of poor clinical response to medical treatment and to rule out abscess formation[5,15].

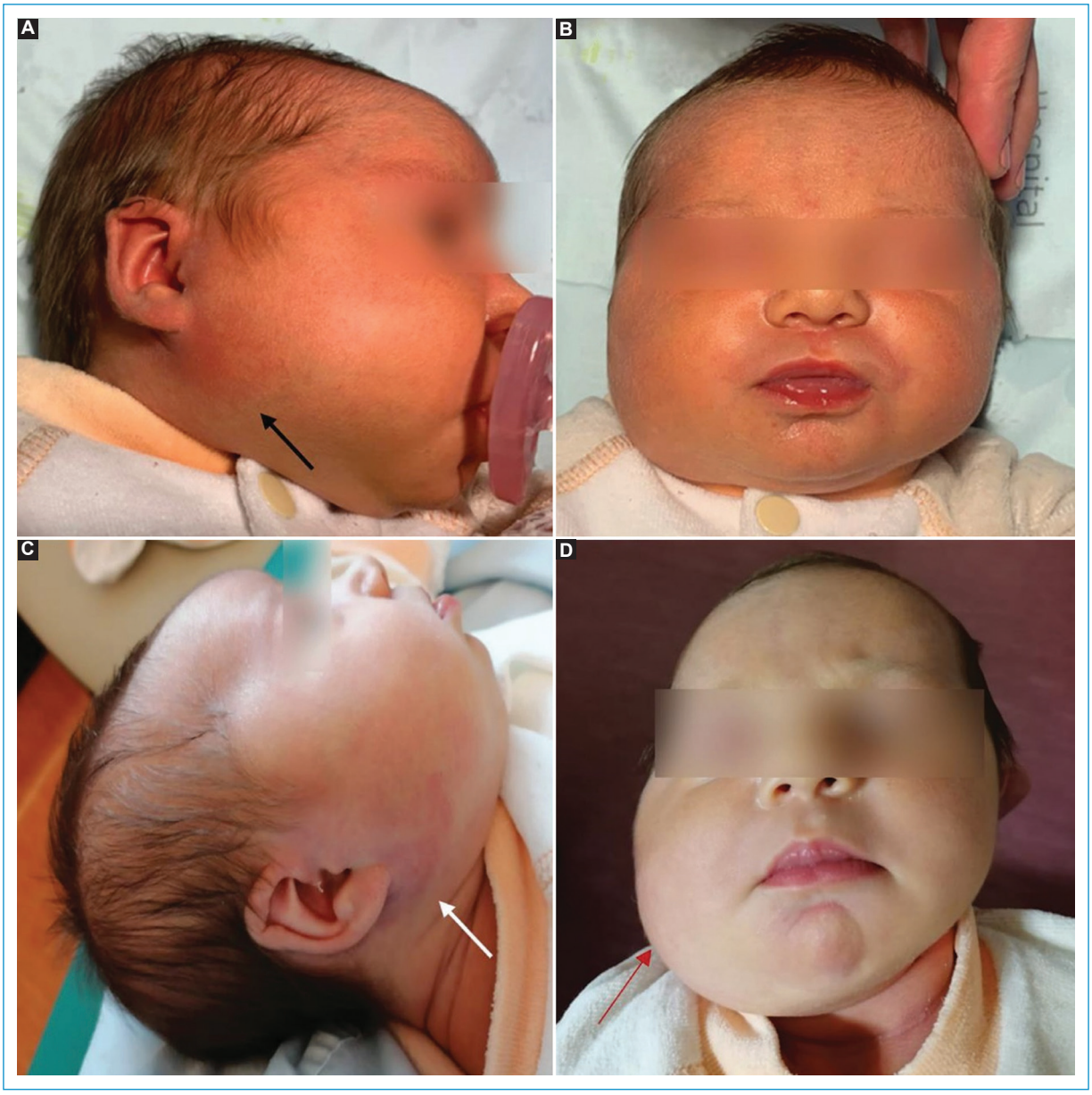

Figure 3. **(A)** and **(B)** presentation five hours after admission: swelling in the right parotid region, local inflammatory signs (black arrow) and asymmetrical face with effacement of the mandibular angle. **(C)** and **(D)** presentation 24 hours after admission: worsening of inflammatory signs (white arrow), in particular, swelling of the parotid region (red arrow).

Although ANP is a clinical diagnosis, in our case, the ultrasound was extremely important since initial findings were subtle and pus drainage occurred afterwards. Laboratory findings were as expected.

Most cases are managed conservatively with antibiotic therapy and adequate hydration[15]. Given the broad bacterial spectrum, most authors recommend empirical treatment with a combination of antistaphylococcal agent and aminoglycoside or third-generation cephalosporin, with adjustment after cultures' results are available[3,7].

The optimal treatment duration is not known, but a minimum period of 7-10 days is usually sufficient[6,9,15] or until the symptoms have resolved[4]. In general, antibiotics lead to clinical improvement within the first 48h with parotid swelling reduction. In rare cases of inadequate response to antibiotics or when organized abscesses are detected, surgical drainage is required[4,7]. In this case antibiotics were adjusted after the recommended minimum time, despite lacking bacterial isolation, due to remarkable clinical improvement.

Currently, due to prompt antibiotic therapy, complications such as salivary fistula, facial palsy, submandibular sialadenitis, mediastinitis, sepsis and meningitis are rare[7,9]. The prognosis of the disease is generally excellent and recurrence is uncommon[3,8]. as in this case.

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Conflicts of interest

None.

Ethical disclosures

Protection of human and animal subjects. The authors declare that the procedures followed were in accordance with the regulations of the relevant clinical research ethics committee and with those of the Code of Ethics of the World Medical Association (Declaration of Helsinki).

Confidentiality of data. The authors declare that they have followed the protocols of their work center on the publication of patient data.

Right to privacy and informed consent. The authors have obtained the written informed consent of the patients or subjects mentioned in the article. The corresponding author is in possession of this document.

References

1. Aguiar T, Dias A, Ferreira G, Machado M do C. Parotidite Bacteriana Neonatal. *Acta Pediatr Port*. 2003;34:17-20.
2. D'Souza JN, Geary C, Mukerji S. Neonatal parotid gland enlargement: Is it suppurative parotitis? a case report. *Am J Case Rep*. 2012;13:41-43.
3. Prior AR, Abrantes M. Parotidite bacteriana bilateral no recém-nascido. *Nascer e Crescer*. 2017;XXVI(1):53-56.
4. Sabatino G, Verrotti A, De Martino M, Fusilli P, Pallotta R, Chiarelli F. Neonatal suppurative parotitis: A study of five cases. *Eur J Pediatr*. 1999;158(4):312-314.
5. Cho JY, Youn JH, Park JS, Cho JM. The First Case of Acute Neonatal Suppurative Parotitis Caused by Escherichia coli in Korea. *Neonatal Med*. 2020;27(2):94-98.
6. Özdemir H, Karbuz A, Çiftçi E, Fitöz S, Ince E, Doğru Ü. Acute neonatal suppurative parotitis: A case report and review of the literature. *Int J Infect Dis*. 2011;15(7):500-502.
7. Ismail EA, Seoudi TM, Al-Amir M, Al-Esnawy AA. Neonatal suppurative parotitis over the last 4 decades: Report of three new cases and review. *Pediatr Int*. 2013;55(1):60-64.
8. Costa L, Leal L, Vales F, Santos M. Acute parotitis in a newborn: a case report and review of the literature. *Egypt J Otolaryngol*. 2016;32(3):236.
9. Decembrino L, Ruffinazzi G, Russo F, Manzoni P, Stronati M. Monolateral suppurative parotitis in a neonate and review of literature. *Int J Pediatr Otorhinolaryngol*. 2012;76(7):930-933.
10. Spiegel R, Miron D, Sakran W, Horovitz Y. Acute neonatal suppurative parotitis: Case reports and review. *Pediatr Infect Dis J*. 2004;3(1):76-78.
11. Megged O, Baskin E. Neonatal Parotitis. *J Pediatr*. 2018;196:319.
12. Avcu G, Belet N, Karli A, Sensoy G. Acute suppurative parotitis in a 33-day-old patient. *J Trop Pediatr*. 2015;61(3):218-221.
13. Singh K. Bilateral parotid abscess in a neonate [4]. *Indian Pediatr*. 2006;43(11):1009-1010.
14. Deepak K, Garima G, Jhamb U. Bilateral acute neonatal suppurative parotitis: A rare finding in neonatal age. *J Neonatal Perinatal Med*. 2015;8(1):63-65.
15. Costa FD, Andrade DR, Cunha FI, Fernandes A. Group B streptococcal neonatal parotitis. *BMJ Case Rep*. 2015;2015:2014-2016.
16. Ujita N, Kawasaki Y, Matsubara K, et al. Late onset group B streptococcus disease manifesting as acute suppurative parotitis. *IDCases*. 2020;21:e00799.
17. Guerra AAH, Osguthorpe RJ. Acute neonatal parotitis caused by streptococcus pyogenes: A case report. *Clin Pediatr (Phila)*. 2010; 49(5):499-501.