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Case report

Bilateral acute suppurative parotitis in a 57-day-old girl: an unusual presentation

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Abstract

Introduction: Acute suppurative parotitis (ASP) is a rare condition in early infancy. It usually presents unilaterally, with inflammatory signs in the parotid region and non-specific systemic symptoms. *Staphylococcus aureus* (*S. aureus*) is the most common causative agent of ASP, and male sex, prematurity, low birth weight, dehydration, parotid duct obstruction and immunosuppression are risk factors. Most reported cases occur in the neonatal period, and they can be complicated by abscesses and sepsis.

Description of case: We present a case of a 57-day-old girl with a history of left submandibular tumefaction, fever, and irritability. Parotid ultrasonography revealed findings compatible with bilateral parotitis and associated phlegmon. Empirical treatment with flucloxacillin was initiated. On the sixth day, the worsening of inflammatory signs on the left side and the onset of inflammatory signs on the right side occurred, and ultrasonography revealed areas of necrotic content bilaterally. Ultrasound-guided drainage of the abscess collections isolated flucloxacillin-sensitive, penicillin-resistant *S. aureus* from the pus culture. Antibiotic treatment was adjusted, and the patient completed 7 days of flucloxacillin and 4 days of gentamicin treatment, with regression of the swelling and complete resolution. No risk factors for ASP were identified. An immunological study was performed, the results of which were normal. During follow-up, no recurrence or complications were reported.

Conclusions: Although ASP is less frequent now than in the past, we must be aware of potentially fatal complications if adequate treatment is not initiated early. Unlike most reported cases, the present case involved an exuberant, bilateral presentation in a girl after the neonatal period, without relevant risk factors, complicated despite the initiation of antibiotic therapy. This

case required the exclusion of immunodeficiency, as otolaryngological presentations are a common manifestation of immunodeficiencies.

Keywords

Suppurative parotitis, abscess, infant, *Staphylococcus aureus*, flucloxacillin, gentamicin.

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Introduction

Acute suppurative parotitis (ASP) is a rare infection in childhood. Classically, it is characterised by parotid gland swelling, pus drainage from Stensen's duct, and growth of pathogenic bacteria in the pus culture. The prevalence of ASP is 3.8-14/10,000 in early infancy [1, 2], with a decline in prevalence noted in recent decades [2]. In 2004, Spiegel et al. reported that only 32 cases were published in the English language in the past 35 years, most of which occurred during the neonatal period [1, 3]. In addition, 16 more cases have been reported since 2004 [4]. In Portugal, the first documented case was an 8-day-old boy [5]. A study in Cambodia revealed an age distribution of cases between 0.7 and 14.6 years, and no neonatal cases were reported [6]. Prematurity is considered a major risk factor for ASP [7]. ASP is more prevalent among males than females, with a ratio of almost 3:1 [1]. Other risk factors include low birth weight, parotid duct obstruction, immunosuppression [1], and dehydration [8]; however, some cases described in the literature do not identify any risk factors [8]. *Staphylococcus aureus* (*S. aureus*) is the most common agent, but other Gram-positive and Gram-negative microorganisms can also be aetiological agents [1, 8, 9]. There are several complications associated with ASP, including abscess and sepsis [8]. Hydration and adequate antibiotic treatment should be initiated early, and surgical drainage may be necessary [8].

Case report

A 57-day-old girl presented with a 1-day history of left submandibular tumefaction, fever, and irritability (**Fig. 1**). There were no changes in food intake; however, some irritability with crying periods was reported by the parents. There was no history of trauma, and the mother denied any recent history of breast tenderness or skin infection. The patient was delivered by caesarean section at 38 weeks and 6 days of gestation, and her birth weight was 3,630 g. Prenatal and family history was unremarkable. Because the recovery of birth weight occurred at 30 days of life, the baby was mixed-fed, and a posterior weight gain of 25 g per day was observed. On admission, she was apyretic and haemodynamically stable. An area of painful and firm left submandibular swelling was noted, clearing the jaw angle, without output of purulent content from the ipsilateral Stensen's duct. Laboratory examination indicated lymphocytic leucocytosis, anaemia, thrombocytosis, and elevated C-reactive protein, and high erythrocyte sedimentation rate. There

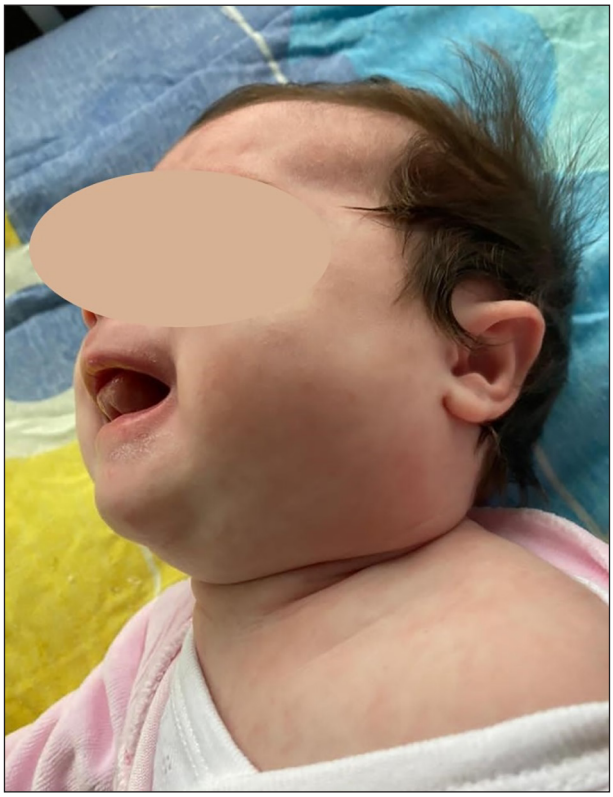

Figure 1. Left submandibular tumefaction at presentation.

were no other relevant changes in biochemical parameters, and serum amylase was normal (**Tab. 1**). The first ultrasonography revealed bilaterally enlarged parotid glands, with a predominance on the left, diffuse heterogeneous structure, and small, reactive lymph nodes close to the tails of both parotid glands. These findings were compatible with bilateral parotitis. Blood and urine cultures were collected. Initially, treatment was deferred, with close clinical surveillance. However, on the second day of admission, due to persistent parotid swelling with maintained apyrexia, ultrasonography was repeated and revealed the presence of a phlegmon in both parotids, with a diameter of 28 mm and 18 mm on the right and left, respectively. Intravenous empirical antibiotic therapy was started with flucloxacillin. On the sixth day, due to the worsening of inflammatory signs on the left side and the onset of inflammatory signs on the right side (**Fig. 2**), ultrasonography was repeated and revealed nodular areas of necrotic content bilaterally (**Fig. 3**). Laboratory tests revealed aggravated inflammatory markers, with increased white blood cell count, thrombocytosis and elevated erythrocyte sedimentation rate (**Tab. 2**). Ultrasound-guided drainage of the abscess collections isolated flucloxacillin-sensitive, penicillin-resistant *S. aureus* in the pus culture, and breast milk culture revealed *Leuconostoc mesenteroides ssp. cremoris* and *Staphylococcus epidermidis*. Blood and urine cultures were negative. The antibiotic therapy was adjusted, and she completed 7 days of flucloxacillin and 4 days of gentamicin treatment, with regression of the swelling and complete resolution. No risk factors for ASP were identified. An immunological study was performed, which was normal. During follow-up, no recurrence or complications were reported.

Table 1. Initial laboratory results.

Haemoglobin	9.4 g/dL
White blood cell count	16,800/mm^3
Polymorphonuclear leukocytes	6,500/mm^3
Lymphocytes	6,600/mm^3
Platelets	456,000/mm^3
C-reactive protein	7.53 mg/dL
Erythrocyte sedimentation rate	51 mm/h
Aspartate aminotransferase	22 IU/L
Alanine aminotransferase	13 IU/L
Amylase	< 4 IU/L

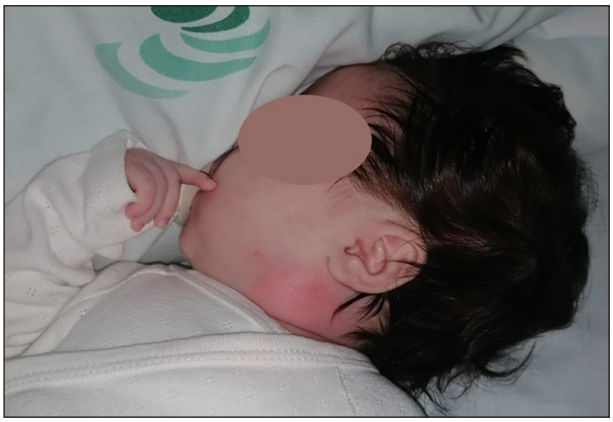

Figure 2. Bilateral submandibular tumefaction with worsening of inflammatory signs.

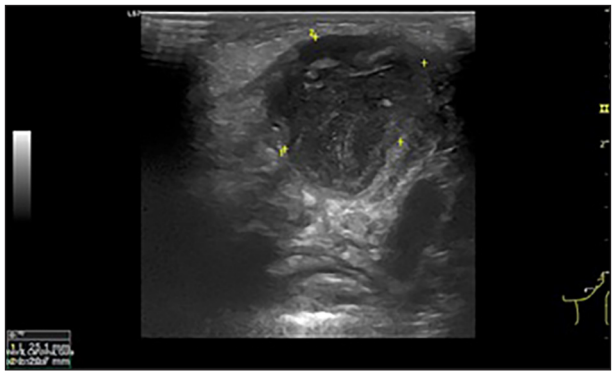

Figure 3. After worsening of inflammatory signs, ultrasound demonstrated echostructural heterogeneity of the parotid glands, with heterogeneous hypoechoic pseudonodular areas in the lower portions bilaterally, with areas of necrosis.

Table 2. Laboratory results after clinical worsening.

Haemoglobin	9.8 g/dL
White blood cell count	28,720/mm^3
Polymorphonuclear leukocytes	11,200/mm^3
Lymphocytes	14,000/mm^3
C-reactive protein	0.27 mg/dL
Erythrocyte sedimentation rate	60 mm/h
Urea	26.0 mg/dL
Creatinine	0.40 mg/dL
Aspartate aminotransferase	28 IU/L
Alanine aminotransferase	20 IU/L

Discussion

ASP presents with low prevalence in the literature. Most reported cases occur in the neonatal period, and prematurity, male gender, low birth weight, parotid duct obstruction, immunosuppression and dehydration are considered risk factors [1, 7, 8]. ASP may develop due to the retrograde flow of oral cavity microorganisms into the parotid gland or rarely due to haematogenous dissemination. The parotid gland is more frequently infected than other salivatory glands because of its

serous secretions that lack bacteriostatic agents [8, 10]. Dehydration and stasis, either due to a decrease in saliva production or congestion in Stensen's duct, are factors that promote the development of ASP [8]. ASP is more common in preterm babies probably due to prolonged hospitalisation, increased bacterial colonisation, trauma to the oral cavity, and dehydration due to nutritional problems [7, 8]. Furthermore, the infection can be attributed to infected breast milk or formula feeding [8]. The present case did not occur during the neonatal period, although we highlight its early occurrence in the post-neonatal period. Prematurity and low birth weight were not present; however, weight gain during the neonatal period was not adequate. There was no history of hospitalisation and no signs of dehydration on admission. However, there was a history of a probable insufficient oral supply during the neonatal period, with significant excess weight loss suggesting dehydration and stasis. Recently, it has been suggested that caesarean section is a risk factor for ASP because the oral flora of newborns is mainly composed of the flora of the mother's skin and opportunistic bacteria from the hospital environment after caesarean section, unlike vaginal delivery [11]. Furthermore, there was no known history of mastitis in the mother; breast milk culture revealed contaminating flora that was assumed to not be causative of ASP.

The diagnosis of ASP is clinical and is commonly characterised by fever and swelling and erythema over the affected parotid gland. Pus exuding from Stensen's duct or aspirated from the affected gland is considered pathognomonic of ASP [9]. Bilateral ASP is uncommon, as 77% of reported cases are unilateral [9]. Laboratory findings in ASP are usually nonspecific, with leucocytosis and neutrophilia reported in 71% of cases [1]. Despite the initial innocuous laboratory findings, our case demonstrated aggravation of inflammatory markers during the first days of hospitalisation, with no clinical response. Spiegel et al. reported that serum amylase levels are elevated in 45% of ASP cases. This finding may be related to the immaturity of this salivary isoenzyme activity in newborns [1]. Pus culture confirms the diagnosis and guides treatment [12].

S. aureus is the most prevalent microorganism in ASP, as in our case, accounting for 55-61% of cases [1, 9]. Methicillin-resistant *S. aureus* (MRSA) is associated with prolonged Intensive Care Unit hospitalisation [13]. Other possible organisms include other Gram-positive and Gram-negative bacteria, such as *Escherichia coli*, *Klebsiella spp.*, and *Pseudomonas spp.*, and anaerobes, such as *Bacteroides spp.*, *Fusobacterium spp.*, and *Mycobacterium spp.* [8, 13]. To the best knowledge of the authors, no cases of *Leuconostoc* parotitis have been described in the literature.

In cases of swelling of the parotid region, differential diagnoses must be considered, such as trauma, lymphadenitis, haemangioma, adenoma, lipoma, parotid gland duct anomalies, intraglandular abscess, or neoplasia [8]. Laboratory examination may not be useful to differentiate between these entities, as inflammatory markers are not usually elevated in most cases of ASP. Therefore, ultrasonography plays an important role in distinguishing these conditions and excluding predisposing factors. In ASP, parotid ultrasound usually demonstrates a diffusely enlarged parotid gland with heterogeneous echo structure and abscess in some cases [12].

Regarding treatment, initially, adequate rehydration must be ensured, which reduces salivary stasis [14]. Empirical antibiotic therapy should be initiated after obtaining pus and blood cultures to cover possible agents. In the literature, a combination of an antistaphylococcal agent with aminoglycosides is suggested as the initial treatment, adding vancomycin if MRSA is suspected [1]. Significant improvement occurs within 24-48 hours in 78% of cases, and complete remission is reported in 83% of cases when appropriate treatment is initiated [1]. Antibiotic therapy should be maintained for 7-10 days depending on clinical response [2, 10, 12, 13]. In case of inadequate response within 48 hours of antibiotic therapy and/or the presence of fluctuance of the gland [2, 7], complications must be considered. In case of an abscess, which is estimated to occur in 23% of cases, surgical drainage should be performed [8]. In our case, ultrasonography was performed due to clinical aggravation during intravenous antibiotic therapy, revealing the presence of a complication.

In modern times, the prognosis of ASP is usually good, in contrast with a higher incidence of complications, such as bacteriemia and sepsis, in the past [1]. Other possible complications are fistula formation, facial paralysis, osteomyelitis of the jaw or temporomandibular joint, jugular vein thrombophlebitis, respiratory obstruction, mediastinitis, septicaemia, and meningitis [8, 14], although these are not frequently reported in the literature.

An immunological study was necessary due to the bilateral and exuberant presentation and the absence of known risk factors. The defence mechanisms of the gastrointestinal tract depend on cellular and humoral immunity and mucosa-specific mechanisms, like mucociliary clearance and immunoglobulin A antibody secretion [15]. Otolaryngological presentations are a common manifestation of immunodeficiencies [15, 16]; however, the immunological study was normal in our case.

In case of recurrence, evaluation for ductal obstruction or sialectasis should be performed [10].

Conclusions

ASP is a rare condition in early infancy; however, it must be considered in the presence of inflammatory signs of the parotid gland, especially with the classic triad of parotid swelling, pus drainage from Stensen's duct, and growth of pathogenic microorganisms in pus cultures. Usually, complete remission occurs with hydration and appropriate antibiotic treatment. Although less frequent nowadays, we must be aware of potentially fatal complications if adequate treatment is not initiated early. Unlike most reported cases, the present case involved an exuberant bilateral presentation in a girl after the neonatal period, without risk factors and complicated despite the initiation of antibiotic therapy. This report required the exclusion of immunodeficiency because otolaryngological presentations are a common manifestation of immunodeficiencies.

Informed consent

Written informed consent was obtained from the patient's legal guardian for the publication of this case report and respective images.

Declaration of interest

The Authors have no conflicts of interest. This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

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