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Acute Suppurative Parotitis in a 22-Day-Neonate with Sepsis: A Rare Case Report

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What is known on this subject?

Acute suppurative parotitis (ASP) in the neonatal period is a rare phenomenon characterized by swelling, pain, and erythema over the parotis gland.

What this case report adds?

Although rare, ASP should be suspected in patients diagnosed with late-onset sepsis with erythematous preauricular mass with or without any predisposing factors.

ABSTRACT

Acute neonatal suppurative parotitis is a very rare condition that causes parotid swelling and purulent exudation into the oral cavity through the Stenson duct. The prognosis is usually excellent, but may be complicated by sepsis, meningitis, and abscesses. A case of sepsis and unilateral suppurative parotitis in a 22-day-old male infant successfully treated with intravenous antibiotics is presented here. Although rare, acute suppurative parotitis can be discovered in neonates, especially in 2-4 weeks of age, and is mostly caused by *Staphylococcus aureus*. Prompt diagnosis, appropriate intravenous antibiotics, and adequate hydration should be initiated immediately to prevent complications.

Keywords: Newborn, parotitis, inflammation, sepsis

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Introduction

Acute suppurative parotitis (ASP) is very rare in the neonatal period, with a prevalence of between 3.8 and 14/10,000 newborn emergency department admissions (1,2). Dehydration, prematurity, low birth weight, immune system defects, trauma, and ductal obstruction are risk factors for neonatal ASP (1,3). Hydration, drawing specimens for pus, blood, and if necessary, cerebrospinal fluid (CSF) cultures, and immediate empirical antibiotic therapy covering possible agents are important in the treatment and prevention of complications (4). Herein, we present the case of a 22-day-old neonate who developed severe late-onset sepsis and right ASP.

Case Report

The male infant was born at 37 weeks of gestation, weighing 3300 g, via normal vaginal delivery to a primiparous 24-year-old mother. Exclusively breastfed neonate was brought to the emergency department at 22 days of age with complaints of high fever, uncontrollable crying, and facial swelling on the right side for the previous 24 h. There was no history of trauma or infection/abscess formation in the mother's nipples or areola. The rectal body temperature was 38.2 °C, respiratory rate was 50 beats/min, arterial blood pressure was 97/66 mmHg, and heart rate was 166 breaths/min. The patient weighed 3200 g and appeared lethargic, dehydrated,

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with decreased peripheral perfusion, depressed neonatal reflexes, and a prolonged capillary refill time of 7 s. The right preauricular region and the right side of the cheek were swollen, tender, and erythematous (Figure 1A). When the right parotid gland was compressed, pus was released into the oral cavity via the Stenson duct (Figure 1B). Laboratory tests revealed a white blood cell count of 18,660/mm^3, 61% neutrophil predominance, hemoglobin level of 14.1 g/dL, thrombocyte count of 254,000/mm^3, and C-reactive protein level of 0.48 mg/dL (normal range 0-0.3). Serum biochemistry was normal, with amylase levels of 4 IU/L (normal range, 28-100 IU/L). Blood gas analysis revealed pH: 7.28, pCO_2: 45.3, lactate: 6.0, HCO_3: 19.4, base excess: 4.4, and urinalysis of the catheterized specimen was normal. The examination of the CSF was normal. Ultrasonographic (US) examination revealed an enlarged, hypoechoic, and hypervascularized right parotid gland with adjacent millimetric lymph nodes (Figure 2A, B). On the second day of treatment, the patient still had signs of severe sepsis and high fever. Magnetic resonance imaging (MRI) of the parotid gland was performed to exclude any abscess formation/congenital tumor or malformations of the venous and lymphatic system that were secondarily infected. It revealed an enlarged gland with no evidence of mass or abscess formation after gadolinium administration (Figure 2C, D). The neonate was diagnosed with ASP and late-onset neonatal sepsis. Warm compresses were applied to the gland, along with intravenous hydration, analgesics, vancomycin, and cefotaxime. After three days of intravenous antibiotics, the patient's condition improved, and the swelling disappeared. Blood, urine, and CSF cultures remained sterile. However, *Staphylococcus aureus* was isolated from the pus culture and was sensitive to vancomycin treatment; therefore, no change in antibiotic treatment was needed.

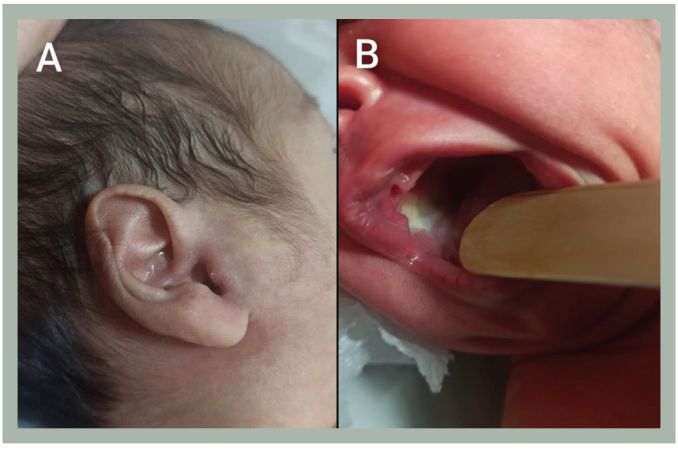

Figure 1. (A) Right facial swelling and erythema. (B) Pus coming from Stenson duct upon massage on parotid gland

Pre-discharge ultrasound of the parotid gland was normal, with improvement of parotid edema. Immunoglobulin levels and lymphocyte subsets were also within age-appropriate ranges. The patient was discharged after 10 days of intravenous antibiotics. The family of our patient provided written informed consent for publication of their child's information.

Discussion

In infants, suppurative infection of the salivary glands is extremely rare, with the parotid gland being the most commonly affected gland (1). Paouris et al. (3) reported only 65 cases of neonatal ASP in the English literature between 1970 and 2020. Prematurity, male gender, dehydration, immune deficiency, trauma, ductal occlusion by tumor, sialolithiasis, breastfeeding in the case of mastitis, or consumption of contaminated formula are risk factors (2,4). Dehydration leads to decreased salivation, impaired salivary flow resulting in salivary stasis, and increased bacterial ascent from the oral cavity to the salivary gland (5).

Infection ascending from the oral cavity through the Stenson duct is the most common route of colonization; hematogenous bacterial germination is less common. The most common organism isolated from pus specimens is *Staphylococcus aureus*. Other Gram-positive microorganisms

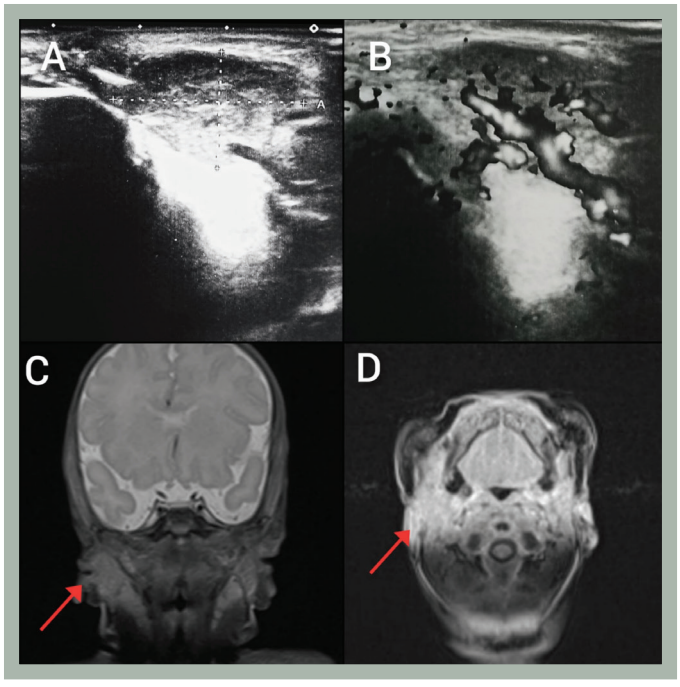

Figure 2. (A and B) Diffuse enlarged (22.3x13.7 mm in diameter), edematous and hypervascularized right parotid gland on ultrasound images. (C and D) Magnetic resonance images with gadolinium administration of the right parotid gland, showing enlarged gland (arrows), with no evidence of mass or abscess formation

such as *Streptococcus viridans, Streptococcus pyogenes,* and *Streptococcus agalactiae,* Gram-negative organisms including *Escherichia coli, Klebsiella, Pseudomonas,* and *Moraxella catarrhalis,* and anaerobic species such as *Bacteroides melaninogenicus, Fusobacterium nucleatum, Peptostreptococcus,* and *Prevotella* spp. can be isolated (4,6,7). Similar to most literature, *Staphylococcus aureus* was isolated from the pus culture of our patient, whereas cultures of blood, urine, and CSF remained sterile.

The diagnosis of the disease is primarily based on the clinical picture, although laboratory tests and imaging studies may also be helpful. Diagnostic criteria are parotid enlargement with or without redness, purulent exudation from Stensen's duct, and the isolation of pathogenic bacteria from pus culture (8). Approximately 50% of neonates with ASP are afebrile, and the condition is typically unilateral, and bilateral involvement is extremely rare. Laboratory tests are typically non-specific. Because of immature salivary isozyme activity in neonates, serum amylase levels are elevated in only 10%-20% of cases (6). The current case was febrile, with impaired peripheral perfusion, depressed neonatal reflexes, and increased lactate on blood gasses; however, no bacteremia or meningitis was detected, laboratory evaluation was non-specific, and the infant gradually improved during the first three days of intravenous antibiotic treatment.

Ultrasound and MRI can be helpful to confirm the diagnosis and exclude other differential diagnoses. US examination, which is non-invasive, cost-effective, and easily accessible, is considered the gold standard for ASP diagnosis. On US, an enlarged and hypervascularized parotid gland, as well as the detection of hypoechoic areas and lymph nodes within and adjacent to the gland, are important indicators of ASP (3). MRI can be used to assist with differential diagnoses and to demonstrate the extent of the disease. In the differential diagnosis, Stenson's duct abnormality or occlusion, sialolith, abscess formation, and parotid gland neoplasms such as hemangiomas, lymphangiomas, lipomas, neurofibromas, and rhabdomyosarcoma should be considered (4).

Although the prognosis tends to be excellent, complications may include abscess formation requiring surgical drainage, facial nerve palsy, septicemia, deep neck infection with mediastinitis, osteomyelitis of the mandible or temporomandibular joint, thrombophlebitis of the jugular vein, meningitis, respiratory distress requiring mechanical ventilation, and even death (5). Antimicrobial treatment should include *Staphylococcus* spp. and anaerobes and Gram-negative spp. Rarely, surgery may be required for abscesses or in patients who do not respond to medical treatment (4,5).

Ethics

Informed Consent: The family of the patient provided written informed consent for publication of their child's information.

Authorship Contributions

Surgical and Medical Practices: M.Ö., D.E., Concept: M.Ö., Design: M.Ö., Data Collection or Processing: M.Ö., D.E., Analysis or Interpretation: M.Ö., D.E., Literature Search: M.Ö., Writing: M.Ö., D.E.

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