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Bilateral Neonatal Suppurative Parotitis With Asymmetric Evolution in an Extremely Preterm Infant: A Case Report

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Abstract

Neonatal suppurative parotitis (NSP) is a rare bacterial infection, usually unilateral and most often caused by *Staphylococcus aureus*. Bilateral involvement is distinctly uncommon, particularly in extremely preterm infants. A female infant born at 26 weeks and six days' gestation (birth weight 1040 g) developed culture-negative late-onset sepsis-like deterioration on day of life (DOL) 13. On DOL 16, bilateral preauricular and submandibular swelling with erythema was noted. Ultrasonography demonstrated bilateral salivary-gland enlargement and hypervascularity without a discrete abscess but could not confidently distinguish parotid from submandibular involvement. By DOL 18, the right preauricular region had spontaneously drained through the skin, while examination of the left side showed induration, warmth, fluctuance, and frank purulence expressed from Stensen's duct. Culture of the purulent drainage grew methicillin-sensitive *Staphylococcus aureus* (MSSA). The infant was treated with intravenous antibiotics, subsequently narrowed to cefazolin, together with warm compresses and gentle gland massage. Bilateral swelling resolved completely by DOL 24 without operative drainage. This case illustrates synchronous bilateral NSP with strikingly asymmetric evolution: active ductal suppuration on one side and spontaneous cutaneous fistulization with decompression on the other. In a deteriorating extremely preterm neonate, direct inspection of Stensen's duct can be decisive when ultrasonography does not clearly localize the involved salivary gland.

Categories: Otolaryngology, Pediatrics, Infectious Disease
Keywords: bilateral parotitis, extreme prematurity, late-onset sepsis, neonatal suppurative parotitis, salivary fistula, staphylococcus aureus

Introduction

Neonatal suppurative parotitis (NSP) is an uncommon but potentially serious bacterial infection occurring during the first weeks of life [1]. Since the first case series of 32 neonates was reported by Spiegel et al. in 2004 [2], cumulative literature reviews now describe more than 90 cases. Most reported cases are unilateral, whereas bilateral involvement accounts for only a small minority of cases. Reviews consistently identify *Staphylococcus aureus* as the predominant pathogen and describe prematurity, low birth weight, dehydration, prolonged hospitalization, reduced salivary flow, and oropharyngeal instrumentation as important predisposing factors [3,4].

The diagnosis is primarily clinical. Purulent material expressed from Stensen's duct is a particularly characteristic finding, while ultrasonography is useful for confirming salivary gland inflammation and identifying a drainable collection [3,4]. Bilateral neonatal disease is especially unusual and may raise concern for viral, systemic, structural, or hematogenous processes. Recent reports have described bilateral parotitis in term infants and sequential involvement of different salivary glands in preterm infants, underscoring the variability of presentation [5,6,7]. If left undiagnosed or untreated, there is a potential for serious complications especially in immunocompromised patients, such as extremely preterm infants due to immature immunity.

Herein, we report a case of methicillin-sensitive *Staphylococcus aureus* (MSSA)-associated bilateral suppurative parotitis with synchronous onset but divergent disease phases in an extremely preterm female infant.

Case Presentation

A female neonate was born at 26 weeks and six days' gestation with a birth weight of 1,040 g following preterm labor. She was admitted to the neonatal intensive care unit for extreme prematurity and respiratory distress syndrome. Respiratory support included non-invasive positive-pressure ventilation. She received total parenteral nutrition through an umbilical venous catheter and subsequently a peripherally inserted central catheter. Empiric antibiotics started at birth were discontinued after 48 hours when blood cultures

remained negative.

On day of life (DOL) 13, the infant developed acute clinical deterioration characterized by recurrent bradycardia, increased oxygen requirement, respiratory distress, abdominal distension, and markedly reduced activity. Blood gas analysis demonstrated severe respiratory acidosis, and she required endotracheal intubation and mechanical ventilation. Blood and urine cultures were obtained, and empiric vancomycin and meropenem were initiated. C-reactive protein (CRP) rose to 219 mg/L. Blood and urine cultures remained negative. The infant improved clinically and was extubated to non-invasive respiratory support on DOL 15.

On DOL 16, examination revealed bilateral preauricular and submandibular swelling with overlying erythema. Doppler ultrasonography of the neck demonstrated bilateral salivary-gland enlargement, increased internal vascularity, and surrounding inflammatory change, without sialolithiasis or a discrete drainable abscess. The sonographic findings did not confidently distinguish parotid from submandibular gland involvement.

On DOL 18, purulent fluid spontaneously drained through the right preauricular skin and was sent for culture. A focused oral examination was then performed. On the left, the parotid region was indurated, warm, erythematous, and fluctuant, and gentle massage produced frank pus from Stensen's duct, confirming parotid involvement. This discharge was not sent for culture as it was assumed to be the same etiology as the contralateral side. The right side showed only mild residual swelling and a small cutaneous punctum at the site of spontaneous drainage, without ongoing discharge (Figure *1*). The overall findings were therefore consistent with bilateral suppurative parotitis at different phases of evolution: an actively suppurative left parotid and a spontaneously decompressed, fistulized right parotid.

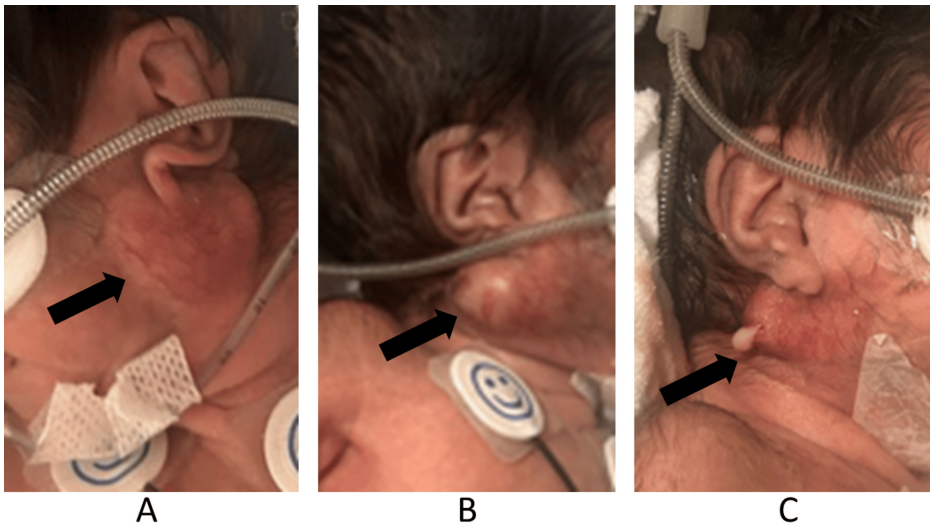


FIGURE 1: Left to right: (A) Left-sided parotid swelling and erythema noted on DOL 16. (B) Right-sided preauricular swelling with overlying erythema noted on DOL 16. (C) Right parotid region following spontaneous decompression, with visible drainage from the site of rupture noted on DOL 18.

DOL: day of life

Parental consent was provided for the use of pictures.

Culture of the right-sided purulent material grew MSSA, and antimicrobial therapy was narrowed to intravenous cefazolin. Gentle left parotid massage expressed additional purulent material, with subsequent resolution of the fluctuance; warm compresses were used as an adjunctive measure. The right-sided fistulous tract healed spontaneously without any specific wound care. CRP decreased to 23 mg/L by DOL 19. Bilateral swelling resolved completely by DOL 24, and the infant completed a 14-day antibiotic course without surgical drainage, which included 10 days of treatment after parotitis was diagnosed.

Because of the unusual bilateral presentation in an extremely preterm infant, an immunologic evaluation was obtained, including immune-cell profiling, neutrophil oxidative burst testing, and immunoglobulin quantification. Evaluation did not identify evidence of an underlying primary immunodeficiency. The infant

had no recurrent infection, was weaned to room air by DOL 58, tolerated full oral feeds, and was discharged home on DOL 68. The cutaneous fistula site healed completely without visible scarring.

Discussion

NSP remains rare. Published reviews describe a male predominance (67-77%), presentation most often at seven to 14 days of life, purulent Stensen's duct drainage in roughly 85% of cases, fever in fewer than half of infants, and unilateral disease in 77-84% of reported infants [3,4,8]. Bilateral disease is therefore a genuinely uncommon presentation.

The parotid is the salivary gland most frequently affected in neonates. Proposed mechanisms include retrograde ascent of oral flora through Stensen's duct and, less commonly, hematogenous seeding [3,4,9]. The retrograde pathway is favored by reduced salivary flow and local instrumentation. Prematurity, low birth weight, dehydration, prolonged hospitalization, nasogastric/orogastric feeding, suctioning, and immature host defenses have all been reported as risk factors [3,4,8,9]. In this infant, extreme prematurity and intensive-care exposure were prominent predisposing factors. Endotracheal intubation occurred three days before the bilateral swelling was recognized and may have contributed to local ductal stasis or oral bacterial inoculation, although a direct causal relationship cannot be established. A relevant comparator is the case reported by D'Souza et al. involving a 27-week infant with unilateral polymicrobial suppurative parotitis [10]. Although both infants were extremely preterm and required respiratory support, the previously reported case lacked the bilateral, phase-divergent evolution observed here.

Staphylococcus aureus is the dominant pathogen in NSP, accounting for the majority of culture-positive cases [3,4]. In contrast to group B streptococcal parotitis, which is commonly bacteremic, staphylococcal NSP may occur with negative blood cultures [3,4]. Our patient's blood cultures remained negative despite a markedly elevated CRP, while MSSA was recovered directly from the purulent parotid drainage on the right side. These findings confirm a localized MSSA salivary-gland infection occurring during the same clinical interval as the culture-negative sepsis-like deterioration but do not establish the route of infection.

The most useful diagnostic feature in this case was the physical examination of the salivary duct orifices. Ultrasonography appropriately identified bilateral salivary-gland inflammation but did not demonstrate a discrete abscess nor the specific gland involved. Expression of purulent material from the left Stensen's duct established the diagnosis. This emphasizes that ultrasonography should complement, rather than replace, targeted intraoral examination in neonatal salivary gland infection [3,4]. In this infant, the anatomical location of the right sided neck swelling was attributed to right parotid involvement, although no direct visualization of drainage from the right Stensen's duct was visualized since a cutaneous fistula had already formed and drained.

The bilateral presentation also broadens the differential diagnosis. Bilateral parotid swelling classically raises concern for a viral parotitis, including mumps. In this case, purulence expressed from the left Stensen's duct and the asymmetric inflammatory findings strongly suggested suppurative parotitis, while growth of MSSA from the purulent drainage confirmed the bacterial etiology. Suppurative disease also tends to appear heterogeneous and hypoechoic on ultrasound, occasionally with microabscesses, in contrast to the homogeneous appearance of reactive swelling, although this distinction was not definitive in our patient [11]. Other considerations included reactive gland enlargement, lymphadenitis, congenital vascular or lymphatic malformations, and submandibular sialadenitis. A previous report described rapid progression from bilateral cervical lymphadenitis to bilateral parotid abscess within hours in a young infant [12], underscoring the potential overlap. The initial sonographic uncertainty was clinically relevant because isolated neonatal submandibular sialadenitis, including bilateral disease, has also been described.

Management consists of empiric therapy covering gram-positive cocci and oral anaerobic flora, including *S. aureus* with attention to methicillin-resistant types, which can reach up to 19%, and then narrow coverage to culture results. Reported courses range from seven to 10 days, typically an initial intravenous course followed by oral step-down, together with hydration and local supportive measures [3,4,8]. Drainage is reserved for a mature abscess, clinical progression, or failure of medical therapy. Ghosal et al. recently described a preterm infant with sequential left parotid and contralateral submandibular abscesses requiring bedside aspiration after inadequate response to antibiotics; that case followed documented *Staphylococcus* epidermidis bacteraemia, consistent with a hematogenous route of bilateral spread [7]. By contrast, our patient's blood cultures remained persistently negative despite bilateral involvement, making hematogenous dissemination less likely and favoring a retrograde ductal route of infection.

The pattern of disease evolution is the principal teaching point of this report. Both parotid regions were swollen on DOL 16, supporting synchronous bilateral involvement. By DOL 18, however, the right side had spontaneously decompressed through the skin, whereas the left remained actively suppurative, with pus expressed from Stensen's duct through gentle massage. It is unclear why the infection progressed differently on each side. Nevertheless, the contrasting findings demonstrate that synchronous bilateral NSP may be at different stages of evolution at the time of specialist assessment. The infection resolved with antibiotics and supportive measures without operative drainage.

Cutaneous salivary fistula is an uncommon complication of suppurative parotitis. In this infant, spontaneous right-sided fistulization provided decompression and healed without intervention. Together with persistent contralateral ductal suppuration, this illustrates that synchronous bilateral NSP may evolve asymmetrically. The combination of extreme prematurity, bilateral onset, divergent local evolution, and spontaneous unilateral decompression is clinically distinctive among published bilateral presentations.

Conclusions

Bilateral suppurative parotitis should be considered in extremely preterm neonates with acute preauricular or upper-neck swelling, even when the two sides appear clinically different. Examination of Stensen's duct may establish the diagnosis when imaging is nonspecific. In the absence of a discrete abscess or clinical progression, culture-directed antibiotics and supportive drainage measures may avoid operative intervention.

Additional Information

Author Contributions

All authors have reviewed the final version to be published and agreed to be accountable for all aspects of the work.

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Disclosures

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