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Neonatal parotid gland enlargement: Is it suppurative parotitis? A case report

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Summary

Background:	Acute suppurative parotitis (ASP) is a rare finding in the neonate. It is commonly caused by <i>S. aureus</i> but other bacterial isolates may be emerging. Effective treatment includes prompt diagnosis, parenteral antibiotics and supportive measures such as rehydration and bimanual gland massage.
Case Report:	This case report describes an extremely premature female infant with a complicated post-natal course who presented with unilateral swelling of the parotid region. Diagnostic workup revealed purulent exudate from Stensen's duct and ultrasound findings consistent with parotitis. Culture of the exudate showed growth of <i>Staphylococcus aureus</i> and <i>Enterococcus</i> species. The patient responded well to a ten-day antibiotic course and supportive measures.
Conclusions:	ASP, though rare, should be considered in the differential diagnosis of a neonatal parotid swelling since early and prompt diagnosis prevents morbidity and complications.
key words:	parotitis • premature birth • <i>Enterococcus</i> spp. • neonate

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BACKGROUND

Salivary gland infections are uncommon in neonates, but when they occur they most commonly involve the parotid gland. Acute suppurative parotitis (ASP) is estimated to occur in less than 4 in 10,000 hospital admissions for newborns [1]. A study by Spiegel et al identified only 32 cases in the English literature over four decades, and found a 72% male prevalence. Most case series show that 35–40% of the neonates affected were born before 37 weeks gestation [2]. However, to the best of our knowledge ours is the first case report describing neonatal ASP in an extremely premature female baby.

Hematogenous seeding of the parotid gland or ascending infection from the oral cavity through Stensen's duct have both been implicated in the development of ASP [3]. *Staphylococcus Aureus* is the bacteria most commonly identified, though *Escherichia coli*, *Pseudomonas aeruginosa*, and group B streptococci have also been isolated. As expected, cultures isolated from our patient grew *Staphylococcus aureus*, but also *Enterococcus* species, which has not been previously reported in the literature.

Several risk factors for the development of ASP have been identified, including low birth weight, oral trauma, immune suppression, and ductal obstruction. Sepsis and malnutrition are also frequently observed in infants with parotitis. Dehydration is another risk factor as it causes salivary stasis leading to bacterial ascent from the oral cavity.

CASE REPORT

A three-week old female infant was delivered at 27 weeks gestation via Cesarean section for prolonged and premature rupture of membranes. Her post-natal course was further complicated by the development of neonatal respiratory distress syndrome (NRDS), she was initially intubated but then was able to be progressively weaned to continuous positive airway pressure (CPAP) therapy. A chin strap was placed in order to minimize mouth breathing during CPAP therapy. At 20 days of life, the patient developed acute onset right parotid swelling and was placed on intravenous vancomycin and cefotaxime, and Otolaryngology as well as Infectious Disease intervention was requested. On exam, the infant was found to have a 3.0×3.5 cm area of induration and erythema overlying the right lateral neck. The right parotid duct area was found to be inflamed, pus was expressed (Figure 1), and sent for cultures. Blood cultures were also drawn.

Complete blood count revealed hemoglobin of 10.1 g/dL, total white blood cell count of $31.8 \times 10^3/\text{mm}^3$ with a left shift (24% segmental neutrophils, 34% bands). Ultrasound of the area showed an enlarged right parotid gland with heterogeneous, hyperechoic echotexture, without evidence of abscess formation, consistent with parotitis.

Based on clinical and ultrasound findings, the patient was diagnosed with right acute suppurative parotitis (ASP). Cultures from Stensen's duct showed growth of *Enterococcus* and methicillin-sensitive *Staphylococcus aureus*. Sensitivity studies showed both organisms were susceptible to vancomycin. Patient was treated with a 10-day course of vancomycin,

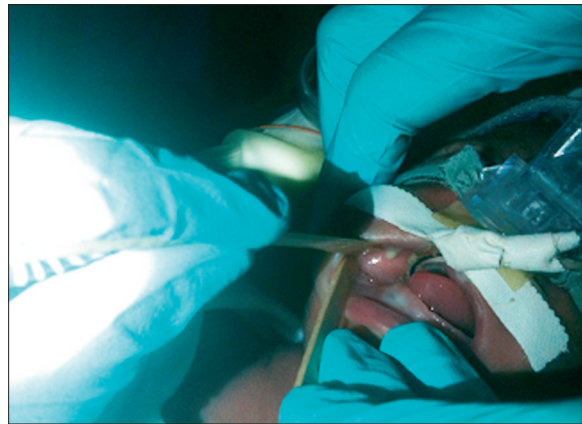


Figure 1. Intraoral view of the right Stensen's duct with expression of purulent discharge.

parotid massage and other supportive measures. The swelling resolved completely without complications.

DISCUSSION

Our patient had many of the risk factors associated with the development of ASP, including low birth weight, extreme prematurity, electrolyte imbalance and neonatal sepsis. Another factor which we feel may have played a role in the development of parotitis was the use of a chin strap to minimize mouth breathing while receiving CPAP therapy.

The diagnosis of ASP is primarily clinical. Sonography of the parotid gland may help confirm the diagnosis and rule out abscess formation. Advanced imaging studies may be considered when the diagnosis is in doubt to rule out other congenital and inflammatory disorders of the parotid gland.

The differential diagnosis of neonatal parotid gland enlargement should include other infectious parotitis, congenital causes such as hemangioma or venolymphatic malformation of the parotid gland and, rarely, tumors and autoimmune conditions. Infectious causes include suppurative parotitis (as was the case in our newborn), mumps parotitis or extrapulmonary manifestations of tuberculosis or human immunodeficiency virus in susceptible populations. Obliteration of Stensen's duct may occur secondary to mucous blockage, sialolithiasis, or even a benign tumor. Autoimmune diseases, such as Sjögren's syndrome, may rarely affect infants [4].

Initial treatment includes aggressive rehydration and electrolyte replacement, reversal of salivary stasis and maintenance of oral hygiene. The sequential management of neonatal parotitis involves a general sepsis work-up, ultrasound of the parotid gland, as well as Otolaryngology and Infectious Disease consults. Given the most frequent pattern of bacterial isolates, empiric antimicrobial therapy is initially focused towards gram-positive bacteria and anaerobes. Beta-lactamase activity has been isolated in over 75% of patients with parotitis, thus penicillins with beta-lactamase activity and antistaphylococcal penicillins or a first generation cephalosporin are generally used for 7–10 days [5]. Regular external bimanual parotid massage starting from the distal end of the gland and working toward the Stensen's duct aid in draining the gland. The need for surgical intervention in neonatal parotitis is extremely rare given the high success rate of medical therapy.

Osteomyelitis of the mandible or temporomandibular joint, thrombophlebitis of the jugular vein, respiratory obstruction and sepsis are potential complications associated with neonatal ASP, and should be considered in the infant with unchanged or worsening symptoms. Facial palsy, salivary fistula and mediastinitis can occur rarely. Mortality is primarily related to the general medical condition of the infant, and is higher in patients who develop complications. However, most patients who receive appropriate parenteral antibiotic therapy recover fully.

CONCLUSIONS

In summary, ASP is uncommon in the neonate. Our case reiterates the fact that a high suspicion for this diagnosis should be maintained in an infant with parotid area swelling who also has predisposing risk factors. Prompt diagnosis leads to early institution of antibiotic therapy and prevents complications. Due to the changing nature of the

bacterial isolates, we emphasize the importance of culture driven antibiotic therapy.

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