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Case report

Neonatal isolated suppurative submandibular sialadenitis: a challenging diagnosis

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Abstract

Introduction: Neonatal isolated suppurative submandibular sialadenitis (NISSS) is uncommon. The first case was described by Shulman in 1950, and since then there have been 39 more cases reported. NISSS is more frequent in preterm neonates in contrast with acute suppurative parotitis, which is mainly present in term neonates. We report a case of NISSS, which highlights the features of diagnosis, treatment, and prognosis.

Case presentation: A premature female was born at 27 weeks and 3 days of gestation from a 22-year-old Caucasian female with good prenatal care. On the 28th day of life, the infant developed suspected sepsis, and on the next day, the infant presented with a red indurated swelling of the left submandibular area. When the oral cavity was examined, spontaneous discharge of pus from the orifice of the left Wharton's duct could be visible. The ultrasound examination confirms the diagnosis of acute submandibular sialadenitis. Bacteriological analysis of the purulent discharge from Wharton's duct isolated heavy growth of methicillin-resistant *Staphylococcus aureus* (MRSA), and the blood cultures were negative. The infant completed 14 days of vancomycin and 10 days of gentamicin.

Conclusion: Although rare, NISSS should not be forgotten as a differential diagnosis in premature infants presenting with submandibular mass with inflammatory signs. Empiric antimicrobial therapy should include *S. aureus* coverage with vancomycin treatment in areas where MRSA is common until sensitivities are known.

Keywords

Neonatal, premature, submandibular sialadenitis, purulent exudation, Wharton's duct, methicillin-resistant *Staphylococcus aureus*.

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Introduction

Neonatal isolated suppurative submandibular sialadenitis (NISSS) is an uncommon condition [1], first described by Shulman in 1950. Since then, there have been 39 additional case reports [2, 3]. Unlike the proclivity for acute suppurative parotitis shown by term neonates, NISSS is primarily confined to preterm births [1, 3].

Prematurity is clearly a major predisposing factor for infection during the neonatal period. However, reliance upon naso- or orogastric tube feedings is a particular risk factor for NISSS [4]. Feeding by tube may reduce salivary secretion to retard or functionally obstruct ductal flow. This leads to stasis, bacterial colonization, and subsequent glandular infection [4, 5]. Dehydration may also render saliva more viscous and slow its flow through Wharton's duct [3, 5].

Herein, we report a case of NISSS, underscoring pertinent diagnostic features, appropriate treatment, and prognosis.

Case presentation

A female infant under our care was delivered prematurely at 27 weeks and 3 days of gestation (birth weight, 755 g). Her 22-year-old Caucasian mother had received good prenatal care, and both parental family histories were negative for serious infectious, metabolic, or neoplastic disorders. The child was delivered by a cesarean section due to preeclampsia, displaying Apgar scores of 5 and 7 at 1 and 5 minutes, respectively. There was respiratory depression at birth, requiring endotracheal intubation and mechanical ventilation until postnatal day 26.

The infant was initially fed parenterally, starting orogastric gavage on postnatal day 3. Neonatal sepsis then developed on day 7, with methicillin-resistant *Staphylococcus aureus* (MRSA) isolated from blood cultures. This necessitated a 20-day course of vancomycin and 14 days of meropenem. Interrupted by sepsis, the enteral feedings resumed on day 15, and parenteral nutrition was discontinued 9 days later.

On postnatal day 28, sepsis was again suspected, signaled by apnea and hyporeactivity. Circulating markers of inflammation/infection, specifically C-reactive protein (64 mg/L) and procalcitonin (1.92 ng/mL), were elevated; and blood cultures were obtained. Intravenous vancomycin and gentamicin were also started as empiric coverage for late-onset sepsis.

The next day, a red indurated swelling (1.5 cm × 2 cm) of left submandibular area was noted. This enlargement was non-mobile, erythematous, and apparently painful. Upon oral inspection, we observed spontaneous discharge of pus from the orifice of left Wharton's duct. The physical exam was otherwise remarkable. The infant remained afebrile and non-irritable, and neither parotid area appeared erythematous, tender, or swollen.

Ultrasound imaging demonstrated a uniformly hypoechoic submandibular gland (**Fig. 1**), with hypoechogenic parenchyma and ductal dilatation (**Fig. 2**). On color Doppler, there was hyperemia (**Fig. 3**). No cysts or calculi were detected. A diagnosis of acute submandibular sialadenitis was made, warranting continuation of antibiotic therapy. Laboratory analysis of the purulent ductal discharge indicated heavy growth of MRSA, whereas blood cultures proved negative.

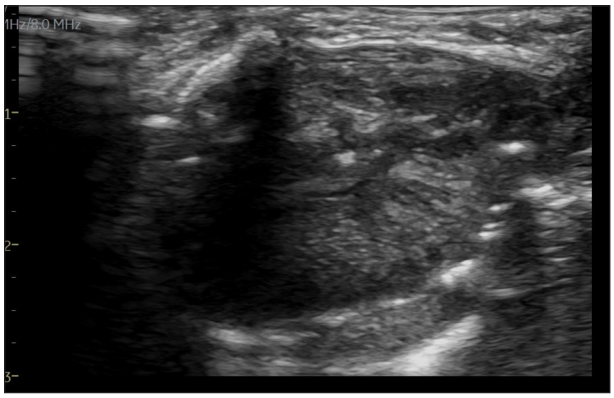

Figure 1. Ultrasound imaging demonstrated a uniformly hypoechoic submandibular gland.

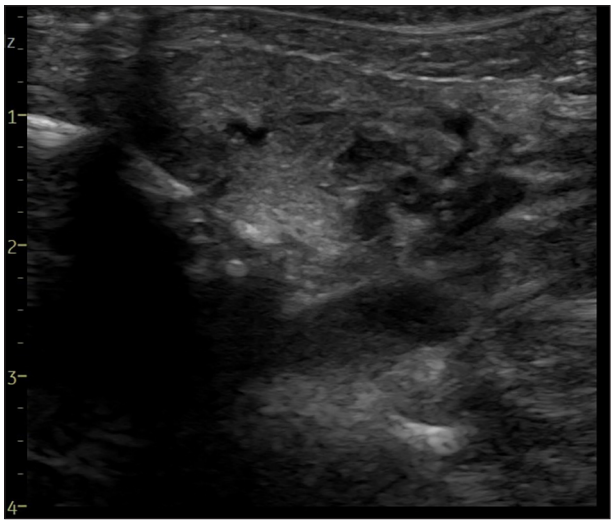

Figure 2. Ultrasound imaging demonstrated hypoechogenic parenchyma and ductal dilatation.

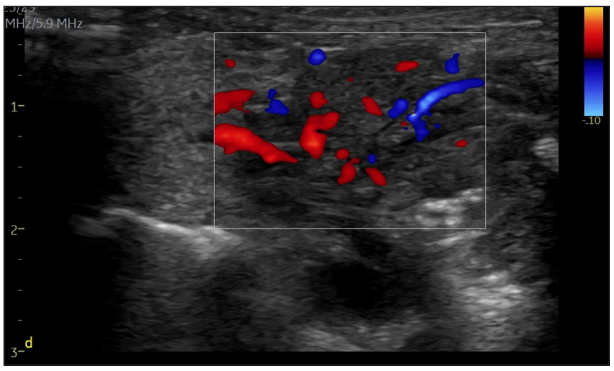

Figure 3. On color Doppler, there was hyperemia.

Another course of vancomycin (14 days) and gentamicin (10 days) was completed. The swelling slowly diminished over a period of 5 days, leaving no residual on follow-up exam. Given her risk factors (i.e., intensive care admission and gentamicin exposure), otoacoustic emissions testing was done prior to discharge, and auditory evoked potentials were later performed, revealing no signs of hearing loss.

Discussion

Submandibular neonatal sialadenitis is characterized by acute onset of submental inflammatory signs [5]. The cardinal element and perhaps diagnostic hallmark is purulent discharge from Wharton's duct (in floor of mouth, below tongue), accentuated by pressure applied to the swollen gland [3]. All diagnostic criteria for suppurative sialadenitis, including submandibular swelling, purulent disharge from Wharton's duct, and pathogenic bacterial growth in cultured exudate, were fully exhibited by our patient.

This infectious disorder must be differentiated from submandibular lymphadenitis, which presents in a clinically similar manner [3, 5]. Other conditions that are easily ruled out would be lymphangioma, teratoma, dermoid cyst, and cystic hygroma. All have distinctive clinical attributes and typically lack inflammatory underpinnings [3].

As we have noted, sepsis was contemplated until the glandular swelling ensued. The fact that blood cultures were sterile makes retrograde spread of oral bacterial flora a more likely explanation than bacteremic hematogenous seeding. In this instance, we believe that factors contributing to development of infection were prematurity, immune compromise, and prolonged orogastric feeding. There was no clinical evidence of dehydration, and ultrasound studies excluded other risk factors, such as congenital malformations (of Wharton's duct or submandibular gland), congenital cysts, or sialolithiasis.

Although *S. aureus* is the one microbe often linked to neonatal sialadenitis, *Streptococci* and Gram-negative bacilli have been implicated as well [6, 7]. Currently, MRSA infection is quite common, so antibiotics effective against MRSA are recommended. Empiric antibiotic treatment should commence as soon as cultures are obtained [6]. The duration of therapy has not been precisely determined, but a minimum of 7-10 days is generally advised or at least a period sufficient to achieve resolution [3]. Appropriate antibiotic coverage and adequate hydration are successful in most cases. If there is clinical worsening after 48 hours of such treatment, incision and drainage are indicated [6].

Conclusion

Despite its rarity, NISSS must be considered in premature infants with submandibular inflammatory swellings. Empiric antimicrobial therapy (in advance of known sensitivities) should include *S. aureus* coverage, using vancomycin in locales where MRSA is common.

Informed consent

Written informed parental consent was obtained to publish this report.

Declaration of interest

None of the Authors have conflicts of interest to declare. There were no funding sources involved, and the corresponding Author was not a research scholarship recipient.

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