CASE REPORT

Isolated suppurative submandibular sialadenitis in a preterm neonate

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SUMMARY

Neonatal isolated suppurative submandibular sialadenitis (NISSS) is rare, with only 21 cases described in the English literature. All cases, with the exception of one, were caused by *Staphylococcus aureus*. In this case report, we describe a preterm neonate with NISSS due to infection by *S aureus* and *Klebsiella pneumoniae* presenting on day 11 of life with submandibular swelling and low-grade fever. Possible contributors for the development of the infection include prematurity, neutropenia, prolonged orogastric feed and bacterial virulence. Antibiotic treatment was effective. To the best of our knowledge, this is the first case report of NISSS caused by a mixed flora.

BACKGROUND

Suppurative sialadenitis is an uncommon infection during the neonatal period and usually involves the parotid gland.^{1,2} Neonatal isolated suppurative submandibular sialadenitis (NISSS) is rare. Only 21 cases have been described in the English literature since its first report by Schulman in 1950.^{3–19} Almost all cases were caused by *Staphylococcus aureus*.

We describe a preterm neonate with NISSS due to infection by *S aureus* and *Klebsiella pneumoniae*. Diagnosis, aetiology and management are discussed.

CASE PRESENTATION

Two male monochorionic diamniotic twin neonates were born to a 29-year-old mother with good antenatal care. Pregnancy was complicated by twin growth discordance and abnormal Doppler findings since 29 weeks' gestation. Caesarean section was performed at 33 weeks' gestation. The mother received two doses of antenatal betamethasone. Apgar scores were 9/9/9 for the first and 9/10/10 for the second twin. Birth weights were 2020 and 1350 g, respectively. Orogastric feed was introduced on day 1 of life. Analytical evaluation excluded twin anaemia-polycythemia sequence on day 1 of life.

The second twin had neutropenia (absolute neutrophil count of 1300/mm³). On day 4 of life, he developed a suspected sepsis, characterised by apnoea and hyporeactivity, and was empirically started on intravenous ampicillin, gentamycin and vancomycin to cover early-onset and late-onset sepsis bacteria. Laboratory tests revealed a total white cell count of 4300/mm³, with a differential of 49% neutrophils and 40% lymphocytes. C reactive protein was 0.42 mg/dL on day 4 and 3.60 mg/

dL on day 5 of life. The blood culture obtained prior to antibiotherapy was sterile. The infant was supported by nasal continuous positive airway pressure from days 4 to 6 of life. Clinical improvement was observed and C reactive protein decreased to 2.37 mg/dL on day 7 of life. On day 11 of life (day 7 for antibiotics), a firm, non-mobile, tender, erythematous mass of about 2.0×1.0 cm was palpable in the right submandibular region. On the day after, a purulent discharge was observed from the orifice of the right Wharton's duct (figure 1). Rupture and spontaneous drainage also occurred through a fistulous cutaneous orifice on the right submandibular region. Physical examination was otherwise unremarkable, with no distress and no signs of dehydration. He had low-grade fever (axillary temperature maximum of 37.8°C) from days 11 to 12 of life. The remaining vital parameters were normal.

His twin brother presented with sepsis by *S aureus* on day 6 of life and was treated with intravenous vancomycin for 10 days. The outcome was favourable.

INVESTIGATIONS

Ultrasound examination on day 12 of life showed an enlarged and heterogeneous right submandibular gland measuring 2.8×1.3 cm, with multiple hypoechoic areas. The left submandibular gland had mild abnormalities. Parotid glands were normal. There was no calcification or gas noted.

Laboratory tests revealed a total white cell count of 17 700/mm³, with a differential of 72%

Figure 1 Right submandibular mass with inflammatory signs and purulent discharge from the orifice of Wharton's duct.

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neutrophils and 21% lymphocytes. C reactive protein was 0.69 mg/dL on day 11 and 2.44 mg/dL on day 13 of life.

Cultures of pus discharge from Wharton's duct and the fistulous cutaneous orifices grew *S aureus* and *K pneumonia*. Both twins were infected by the same strain of *S aureus*, sensible to oxacillin and vancomycin and resistant to penicillin.

DIFFERENTIAL DIAGNOSIS

- ▶ NISSS
- ▶ Submandibular lymphadenitis
- ▶ Lymphangioma
- ▶ Haemangioma
- ▶ Congenital tumours

TREATMENT

On day 12 of life, ampicillin was discontinued. Intravenous vancomycin and gentamycin were empirically maintained. On day 15 of life, gentamycin was replaced by intravenous amikacin, according to the *K pneumonia* antimicrobial susceptibility pattern.

The infant completed 15 days of vancomycin and 8 days of amikacin.

OUTCOME AND FOLLOW-UP

The submandibular swelling was resolved by day 16 of life. The white cell count and C reactive protein were normalised. The infant did not present further evidence of infection and was discharged on day 30 of life.

DISCUSSION

The diagnostic criteria for suppurative sialadenitis, originally described for neonatal suppurative parotitis,² were fulfilled in our case report: submandibular swelling, purulent exudation from Wharton's duct and growth of pathogenic bacteria in pus culture.

As observed in our case report, NISSS typically manifests at the second week of life with a unilateral submandibular swelling associated with inflammatory signs.^{17 18} Low-grade fever and non-specific laboratory findings of leucocytosis with neutrophil predominance are also frequent. Differential diagnosis includes submandibular lymphadenitis that may have similar clinical presentation.^{12 13} Lymphangioma, haemangioma and congenital tumours should also be considered, but they are less likely since signs of inflammation are usually absent, unless there is a secondary infection. On the other hand, haemangioma and congenital tumours are often present at birth, contrary to our case report that only exhibited a submandibular swelling on day 11 of life. History, physical examination and ultrasound examination ruled them out.

The oral cavity is the probable portal of entry, but haematogenous seeding has been described.^{1 18} There are predisposing conditions for NISSS. Previous reports have suggested that prolonged orogastric or nasogastric feeds may result in decreased saliva production and ductal clearance, leading to functional ductal obstruction and local inflammation.^{6 13} In addition, hypostimulation causes bacteriostatic mucus production to decrease and may contribute to infection.¹⁸ Dehydration is also a common predisposing condition by producing salivary stasis.¹

Prematurity has been pointed out as the main risk factor for developing NISSS.^{13 17 18} Preterm neonates usually have a relative immaturity of the immune system, which, along with bacterial colonisation of the oral cavity, prolonged orogastric or nasogastric feeds and increased risk of dehydration, may contribute to the pathogenesis of NISSS.

In our case report, a suspected sepsis preceded NISSS by 7 days. Although the blood culture was sterile, the C reactive protein increased accordingly and the infant did improve after antibiotic initiation. We decided to empirically cover early-onset and late-onset sepsis bacteria because symptoms started on day 4 of life. Early-onset sepsis is usually acquired vertically from the mother and antibiotic coverage should initially be directed against most likely highly pathogenic bacteria, especially group B *Streptococcus* and *Escherichia coli*.²⁰ Late-onset sepsis has been attributed to nosocomial or horizontal acquisition and is mostly caused by coagulase-negative staphylococci.²¹

Bacteraemia and NISSS caused by haematogenous dissemination seem unlikely since blood culture was sterile and clinical manifestations of NISSS only appeared by day 7 of successful treatment with antibiotics. We hypothesise that descending bacteria from the oral flora caused the infection of the submandibular gland. Prematurity, a compromised immune system (neutropenia present at birth), and prolonged orogastric feed may have contributed to the development of NISSS in our case report. No clinical evidence of dehydration was noted. Ultrasound examination excluded other risk factors, such as congenital malformations of Wharton's duct and submandibular gland, congenital cysts and sialolithiasis.

Cultures of the blood and drainage from the involved gland should be obtained. The most frequently found causative bacteria in neonatal sialadenitis is *S aureus*,^{2 17} but streptococci and Gram-negative bacilli such as *E coli*, *Pseudomonas aeruginosa* and *K pneumoniae* and mixed aerobic-anaerobic infections have also been implicated.² In our case report, submandibular gland pus obtained from two distinct locations grew *S aureus* and *K pneumonia*, making contamination unlikely. Both twins were probably colonised by the same strain of *S aureus*. Resistance of *K pneumonia* to gentamycin may have been an important virulence factor to the establishment of the infection. To the best of our knowledge, this is the first case report of NISSS caused by a mixed flora. *S aureus* is the single causative agent in almost all cases of NISSS described, except one that was attributed to *Paeruginosa*.⁷

The mainstay in the treatment of NISSS is the appropriate selection of antibiotics. Empiric antibiotics should be started as soon as cultures are obtained and should cover *S aureus* and Gram-negative microorganisms.¹⁸ Antibiotic sensitivity when available allows adjustment of therapy. In our case report, NISSS developed after 7 days of use of large spectrum antibiotics. We discontinued ampicillin since vancomycin and gentamycin already covered *S aureus* and Gram-negative bacteria. Vancomycin could have been substituted by oxacillin, according

Learning points

- ▶ Neonatal isolated suppurative submandibular sialadenitis is rare but must be kept in mind when evaluating preterm neonates with prolonged orogastric or nasogastric feeds that present with submandibular swelling.
- ▶ Ultrasound examination is a useful diagnostic tool that allows the exclusion of anatomical predisposing abnormalities and the identification of abscess formation.
- ▶ The most common aetiological agent is *Staphylococcus aureus*, but Gram-negative bacilli may also be implicated. Empiric antibiotics should be started as soon as cultures are obtained.

to the *S aureus* antimicrobial susceptibility pattern. Ultrasound examination should be repeated in the absence of clinical improvement after 24–48 h of antibiotics, to exclude abscess formation that may require surgical management.^{10 16–18} In our case report, antibiotic treatment alone was effective. Resolution usually ensues within 1 week^{13 18} and no recurrences have been reported on follow-up.^{17 18}

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