Neonatal suppurative submandibular sialadenitis

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Neonatal sialadenitis of the submandibular gland is a very rare clinical entity. Information about the etiopathogenesis and management of the disease is very limited. Prematurity, prolonged gavage feeding and dehydration are the frequent causes. This report presents a rare case of isolated suppurative submandibular sialadenitis in a full-term newborn without any risk factors. Possible etiology, diagnosis and management of this uncommon disease are discussed.

Key words: newborn, submandibular, sialadenitis.

Infections of the salivary glands are uncommon in the neonatal period and most often involve the parotid gland. Submandibular sialadenitis is exceptionally rare in neonates. It usually follows infection of the parotid gland. Only a few cases of isolated submandibular gland infection without the involvement of the parotid gland have been reported[1-3]. Early diagnosis and antibiotic treatment of suppurative submandibular sialadenitis may prevent complications such as abscess formation, septicemia and respiratory failure. Here, we describe a newborn who developed an isolated submandibular sialadenitis. The infection resolved after the antibiotic therapy.

Case Report

A 17-day-old female neonate presented with a two-day history of swelling and redness of the submandibular region. She was born at 36 weeks of gestation with a birth weight of 2575 g. The pregnancy was uncomplicated. The infant was breastfed and given mother's milk with bottle a few times. The mother had nipple cracks but no signs of mastitis.

Her weight was 2700 g on the day of admission. A firm, tender, erythematous swelling about 2x1 cm in diameter was noticed at the right submandibular region (Fig. 1). There was no evidence of erythema, swelling or tenderness in either parotid region. The infant was afebrile. Purulent exudate exuded from the Wharton's duct when pressure was applied to the gland (Fig. 2). Her vital parameters and hydration status were normal. Her examination was otherwise unremarkable.

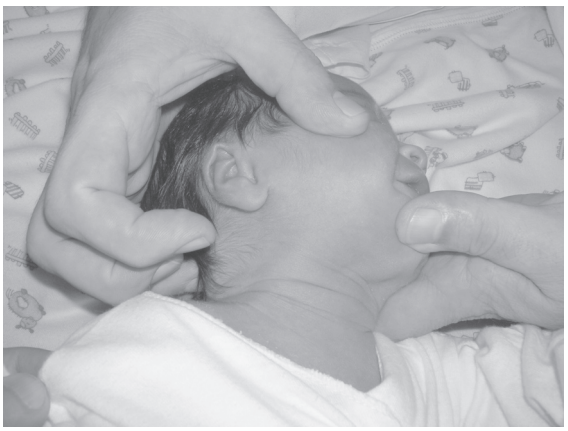

Fig. 1. Neonate with submandibular swelling.

Fig. 2. Purulent exudation from the Wharton's duct.

On admission, the total white blood cell count was 19,000/mm^3 (64% neutrophils, 26% lymphocytes, 6% monocytes, and 4% stab).

Erythrocyte sedimentation rate was 10 mm/hr and C-reactive protein level 2.75 mg/dl. Serum amylase was not increased.

Ultrasound examination demonstrated a generalized swelling, increased vascularity and parenchymal heterogeneity of the right glandula submandibularis without signs of an abscess. No abnormalities of the left submandibular gland or parotid gland were seen (Fig. 3).

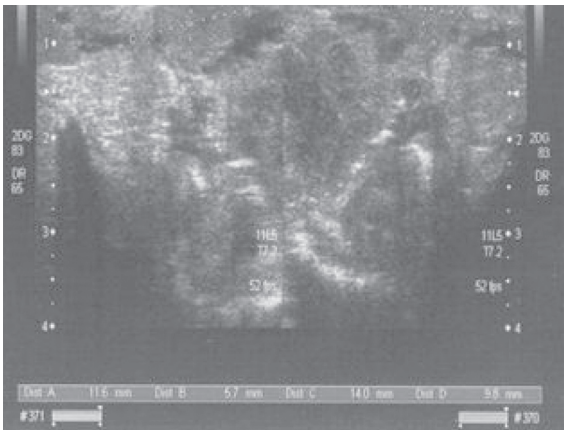

Fig. 3. Oblique axial sonograms of both submandibular glands show increased gland size and mild heterogeneity on the right.

Gram stain of the pus manually expressed from Wharton's duct showed gram-positive cocci. A diagnosis of acute suppurative sialadenitis was established. After the pus and blood cultures were obtained, empiric intravenous treatment with ampicillin-sulbactam (150 mg/kg/d) and gentamicin (7.5 mg/kg/d) was initiated. *Staphylococcus aureus* was isolated from the purulent discharge whereas the neonate's blood cultures and mother's milk were sterile. After the second day, ampicillin-sulbactam (150 mg/kg/d) alone was continued intravenously for another 7 days, during which the palpable mass resolved to 0.5x0.5 cm in diameter. She was discharged with oral amoxicillin/clavulanate for seven more days. Follow-up examination demonstrated no residues or abnormalities of the gland.

Discussion

The first case of neonatal suppurative submandibular sialadenitis was reported by Schulman in 1950[1], and only 19 other cases have been reported since then[2,3]. Submandibular sialadenitis usually follows infection of the parotid gland; submandibular gland infection without involvement of the parotid gland is seen infrequently. Infection of the submandibular gland is rare compared to the parotid gland because it produces more mucus, which is bacteriostatic, protecting the gland from infection[4].

The predisposing factors for suppurative sialadenitis in newborns are prematurity, dehydration, prolonged orogastric feeding and congenital anomalies of the floor of the mouth[5]. A clear association between prematurity and suppurative sialadenitis has been shown previously[6]. Seventy-six percent of neonates with submandibular sialadenitis were born prematurely (≤35 weeks). Prematurity has been described as the main risk factor for developing submandibular sialadenitis, and only three full-term babies with submandibular sialadenitis have been reported previously[3].

Dehydration and gavage feeding have been proposed as other predisposing factors[5], but neither of these was present in our case. Although dehydration has been implicated as a risk factor, signs of dehydration may not be observed. The association has been shown but only two of the cases presented with signs of dehydration[5]. Only a few neonates with submandibular sialadenitis were not gavage-fed[3]. Transmission of bacteria during breastfeeding or through contaminated formula can be a potential cause of sialadenitis. In the presented case, bacterial colonization of the bottle-fed mother's milk with S. aureus could have occurred during storage or warming before feeding. The infant was given breast-milk with a bottle a few times, but the mother had no signs of mastitis and no bacterial growth was observed in her milk.

Staphylococcus aureus is the usual causative organism in neonatal sialadenitis. The other isolated organisms have included streptococci, *Pseudomonas aeruginosa, Escherichia coli* and *Moraxella catarrhalis*. Anaerobic bacteria have been recovered from salivary gland infections in older children and adults[7], but Prevotella species (intermedia/melaninogenica), *Fusobacterium nucleatum*, and *Peptostreptococcus magnus* have recently been reported in two newborns[2]. Suppurative sialadenitis due to methicillin-resistant *S. aureus* (MRSA) has also been described[5]. Polymicrobial aerobic-anaerobic infection is common in these infections. Although all previously reported cases except one[5] were associated with negative blood cultures, hematogenous spread needs to be considered.

The diagnosis of submandibular sialadenitis can be made on clinical grounds. However, systemic manifestations may be minimal in neonates with salivary gland infection[4]. The temperature elevation may be slight and the infants may continue to feed well[9].

Examination with ultrasound is non-invasive, cheap and useful for diagnosis, differential diagnosis and excluding the other predisposing factors like anatomical abnormalities of Wharton's duct, mechanical salivary duct obstruction secondary to a sialolith and infection related to a submandibular gland neoplasm; however, none of the reported infants had oral cavity cysts or calculi.

The administration of antimicrobial therapy is an essential part of the management of patients with suppurative sialadenitis. Most cases respond to antimicrobial therapy; however, sometimes abscess formation requires surgical drainage[8,9]. Empirical antibiotics for sialadenitis in the newborn should cover both gram-positive and gram-negative organisms. Although *S. aureus* is the most common responsible organism in neonatal sialadenitis, *Escherichia coli*, *Pseudomonas aeruginosa*, and *Neisseria catarrhalis* have been reported as the other causative agents[5]. Suitable empiric antibiotic treatment consisted of an antistaphylococcal β-lactam agent and an aminoglycoside given intravenously to neonates until the causative organism was isolated. A penicillinase-resistant penicillin or a first-generation cephalosporin is generally adequate coverage for *S. aureus* infection. However, infection with MRSA may require the use of vancomycin, teicoplanin or linezolid. Clindamycin, cefoxitin, imipenem, or the combination of metronidazole and a macrolide or of a penicillin plus a beta-lactamase inhibitor should provide adequate coverage for anaerobic as well as aerobic bacteria[7].

In conclusion, although isolated neonatal suppurative sialadenitis is rare, it should be suspected even in full-term infants presenting with an erythematous submandibular mass without any predisposing factors.

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