CASE REPORT

Neonatal suppurative sialadenitis: an important clinical diagnosis

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SUMMARY

Neonatal suppurative sialadenitis is a condition causing infection of the salivary glands, most frequently caused by *Staphylococcus aureus*. The vast majority of cases reported in the literature have been in infants with recognised risk factors. This report presents two cases of neonatal sialadenitis in siblings, neither of whom had any predisposing characteristics. The aetiology, diagnosis and treatment of this condition are discussed. This report will also highlight the need for awareness of this condition and its inclusion in considered differentials for neck swelling in all infants.

BACKGROUND

Neonatal suppurative sialadenitis (NSS) is an infection of the salivary glands, often caused by *Staphylococcus aureus*,¹ with a reported incidence of 1 in every 10 000 admissions.² Diagnosis of NSS is performed clinically; features are fever, preauricular or submandibular swelling, and pus exudate at the opening of Wharton's or Stenson's duct.¹ Risk factors include prematurity, prolonged nasogastric feeding, supported ventilation, dehydration and maternal mastitis while breastfeeding.¹⁻³ Treatment of sialadenitis is with intravenous antibiotics; this route has been shown to be important in preventing abscess formation.³ The vast majority of infants make a full recovery and few cases recur.² There is a risk that the sialadenitis can progress to form an abscess,³ or even go on to obstruct the infant's airway, requiring intubation, or the infection could spread to the patient's bloodstream.² The importance of this condition lies in the combination of its seemingly innocuous presentation paired with severe complications when not treated. This report presents two cases of NSS in siblings. The first case is of a male infant with neonatal suppurative submandibular sialadenitis and the second case is of his younger sister, who was diagnosed with neonatal suppurative parotid sialadenitis a few years later. Both of these children went on to recover with no sequelae of the condition, owing to swift diagnosis and initiation of treatment.

CASE PRESENTATION

The first case was a 20-day-old male who was born at term, bottle fed and feeding well. He presented with swelling on both sides of his neck, fever, drowsiness and one episode of vomiting. On examination a 2 cm non-tender submandibular mass was noted on the left side.

INVESTIGATIONS

A full septic screen was performed; blood results showed a raised white cell count and C reactive protein. A sample of the pus pooling at the opening of Wharton's duct was sent for microscopy, sensitivity and culture. This later showed growth of *S. aureus*, sensitive to flucloxacillin and erythromycin. Ultrasound scan supported this diagnosis and confirmed the absence of sialolithiasis.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis should include lymphadenitis, haemangiomas and venous malformations.

TREATMENT

The patient was treated in hospital with intravenous cefotaxime 200 mg three times daily and flucloxacillin 200 mg three times daily, initiated on day 2 and continued for 3 days until discharge.

OUTCOME AND FOLLOW-UP

The child was discharged on day 4 with oral flucloxacillin 100 mg four times daily and intravenous ceftriaxone 200 mg daily. He received this antibiotic regime for 4 days, making the total length of antibiotic therapy 7 days. On day 8 he attended the outpatients' department having made a full recovery.

CASE PRESENTATION

A number of years later, his younger sister presented to the hospital, aged 16 days. Her birth history was unremarkable, she was bottle fed and feeding well. She presented with fever and an erythematous swelling over her left cheek and jaw, around 3 cm in diameter. Some possible swelling of the upper left gingiva was noted.

INVESTIGATIONS

Blood results showed raised white cell count and C reactive protein. Blood cultures grew *S. aureus*, sensitive to flucloxacillin and erythromycin. Mouth swabs grew *S. aureus*, again sensitive to flucloxacillin and erythromycin. An ultrasound scan reported a left parotid swelling but no evidence of abscess.

DIFFERENTIAL DIAGNOSIS

The clinicians' main differentials were parotitis, lymphadenitis or sepsis.

TREATMENT

While an inpatient, the child was treated with intravenous cefotaxime 165 mg three times daily for 6 days.

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OUTCOME AND FOLLOW-UP

The patient was discharged on day 6 with a 5 day course of oral flucloxacillin 75 mg three times daily. At community review on day 10 there were no concerns reported.

DISCUSSION

It is of interest to note that neither of these children had any of the known risk factors for sialadenitis. We explored various possibilities in these children. Some sources have reported that contamination of formula milk could lead to sialadenitis but this seems unlikely as a cause for two distinct cases.⁴ Another proposed cause is oral infection ascending into the salivary duct.¹ This could be a contributory factor in the second case, as some erythema of the gingiva was noted, but even in this instance it was not confirmed as gingivitis, and cannot account for her brother's infection. We considered the possibility of a genetic link as it is unusual for two siblings to present with this same condition. We have carried out a literature search to look at any previous reports of genetic links and we could not find any reported associations. We referred the family to a geneticist in a tertiary hospital, who suspects that there is an underlying genetic basis for this and that it is likely autosomal dominant. Consequently, the two patients have been recruited into the MAGIC study (Manchester Gene Identification Consortium Study).

Although this condition is not common, awareness of sialadenitis is incredibly important. One reason for this is the serious complications that can arise from it should it be left to progress. In 1970, 1/3 of the children in the reported cases died due to the infection⁵; of course, the mortality is far lower now, but these sombre statistics serve as a grave reminder of the possible consequences of not acting quickly and appropriately.⁶ Further to this, the risk of airway obstruction and septicaemia must also be considered.^{7 8} As the blood cultures from the female infant in case 2 came back having grown *Staphylococcus aureus*, it is easy to see how a subclinical septicaemia could be missed, and had she not received antibiotics immediately, the outcome could have been far less favourable.

Posing a threat to swift diagnosis is that sialadenitis often presents a similar clinical picture to other diseases, which have very different treatment courses. For example, mumps can also present with non specific fever, malaise and a preauricular or submandibular mass. Again, similarly to sialadenitis, the diagnosis is usually made clinically. The vital difference is that mumps, being a viral infection, is not treated with antibiotics and is usually managed in the community. In the event of a misdiagnosis, this would mean delaying antibiotic treatment and falsely reassuring parents that the infection is self-limiting, which could lead to progression of sialadenitis and adverse complications.⁹

When sialadenitis has been diagnosed, it must also be ensured that blood culture samples are taken to find the causative organism. Although the majority, 55–87%, of cases are due to *S. aureus*, it has been noted that other causes are more likely to lead to sepsis, possibly due to inappropriate choice of antibiotic.^{2 5 10} Brook suggests that the low diagnosis rate of sialadenitis caused by organisms other than *S. aureus* may be due not to a low rate of incidence, but, in fact, as a result of poor collection and cultivation methods of blood cultures. Evidence this paper gives in support of this theorem is that anaerobes are far more common in the normal oral flora, outnumbering

aerobes by 10–100 times.¹¹ Therefore cultures should be taken without delay and in any situation where sialadenitis does not make a rapid improvement following commencement of antibiotics, repeat cultures should be taken to reassess the culpable organism and its antibiotic sensitivity. Thankfully, in the two cases reported above, both infants made a recovery within days of treatment being started. It must be noted that the antibiotics given were via an intravenous method initially, which has been shown to reduce the risk of abscesses forming when compared with oral antibiotic treatment alone.²

Learning points

- ▶ Although sialadenitis is a rare condition, it should always be considered at least as a differential for an infant presenting with fever and neck or facial swelling; a misdiagnosis poses the risk of serious complications.
- ▶ These case reports demonstrate that it is not essential to have any currently recognised risk factors to develop sialadenitis.
- ▶ Neonatal sialadenitis may have a genetic link and this possibility should be investigated in future cases.
- ▶ Where sialadenitis is suspected, a full septic screen should still be carried out, to check for a concurrent septicaemia and other sources of fever.
- ▶ Intravenous flucloxacillin or erythromycin are the most appropriate antibiotics.

Contributors BP was the consultant in charge of care of the two patients presented in this manuscript. RFR performed a literature review and compiled the case reports from the patient notes.

Competing interests None declared.

Patient consent Obtained.

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