Case report

Late onset group B streptococcus disease manifesting as acute suppurative parotitis

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\textbf{A R T I C L E   I N F O}

\begin{tabular}{l}
Article history: \\
Received 22 April 2020 \\
Received in revised form 4 May 2020 \\
Accepted 5 May 2020 \\
\end{tabular}

\begin{tabular}{l}
Keywords: \\
Acute suppurative parotitis \\
Group B streptococcus \\
Bacteremia \\
Neonate \\
Late onset disease \\
\end{tabular}

\textbf{A B S T R A C T}

Few patients with acute suppurative parotitis (ASP) due to group B streptococcus (GBS) have been documented. Limited data on clinical and microbiological features and infectious route are available. We present a 21-day-old boy with invasive GBS disease manifesting as ASP. The patient was admitted because of irritability, fever, and erythematous swelling over the right parotid area. No purulent material exuded from the Stensen’s duct. Ultrasonography and computed tomography of the neck showed findings indicative of ASP. On the day after admission, blood culture yielded GBS. The isolate was determined as GBS serotype Ia and sequence type-23, and the patient was successfully treated with intravenous ampicillin for 10 days. A review of the literature revealed 11 GBS ASP infants including ours with age at onset between 13 days and 12 weeks. All infants had bacteremia while pus from the Stensen’s duct was detected in only one case. This finding remarkably contrasts with ASP caused by pathogens other than GBS, where the infection usually spreads via a retrograde route from Stensen’s duct. The present case and literature review indicate GBS ASP primarily arises from bloodstream infection, and that ASP should be included in an infectious focus as late onset GBS disease.

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\textbf{Introduction}

Acute suppurative parotitis (ASP) in children is a rare disease, characterized by fever, parotid swelling and tenderness with purulent discharge from the Stensen’s duct \cite{1,2}. Staphylococcus aureus is the most frequently isolated pathogen, but other organisms can be implicated, including streptococci, Gram-negative bacilli and anaerobes \cite{2,3}. We encountered a case of late onset group B streptococcus (GBS) disease manifesting as ASP. We reviewed the literature on ASP caused by GBS in infants, and delineated the clinical and microbiological features and infectious route of this uncommon disorder.

\textbf{Case report}

A 21-day-old male visited our hospital with an 8-h history of irritability and reduced feeding, and 3-h history of fever and swelling of the right periauricular region. The patient was born at 38 weeks gestation by cesarean section because of diminished fetal pulse rate during labor. Apgar scores were 8 and 9 at one and five minutes, respectively, and birth weight was 2933 g. There had been no prolonged rupture of membranes. Culture of the mother’s vagina at 36 weeks gestation was negative. Postnatal courses were unremarkable and the baby was breast-fed after birth.

On presentation, the baby was irritable and looked unwell. Body temperature was 38.6°C, heart rate 162/min, respiratory rate 40/min, and blood pressure 114/71 mmHg. Physical examination showed erythematous swelling over the right parotid area, which rapidly extended to the right submandibular region within 2–3 h. No purulent material exuded from the Stensen’s duct. Laboratory results included white blood cell count 4300/\textmu L, hemoglobin 12.7 g/dL, C-reactive protein 1.3 mg/dL, and a serum amylase concentration 15 IU/L. Cerebrospinal fluid (CSF) examination showed no pleocytosis. On admission, contrast-enhanced computed tomography (Fig. 1) of the neck demonstrated enlargement of the right parotid gland with swelling of adjacent soft tissues, but no abscess formation. Ultrasound examination also revealed unilateral enlarged parotid gland with hypoechoic areas and increased vascularity (maximum diameter, right: \(39 \times 28\) mm; left: \(23 \times 18\) mm).

Based on these findings, a diagnosis of ASP was made. After cultures were obtained from blood, urine, and CSF, the newborn

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was admitted and received intravenous ampicillin (50 mg/kg/dose every 8 h) and cefotaxime (50 mg/kg/dose every 8 h). On the day after admission, the blood culture, but not urine and CSF culture results, yielded *Streptococcus agalactiae*. The treatment was changed to ampicillin alone. On hospital day 3, his fever subsided and local inflammatory signs improved. The patient was discharged without any sequelae after 10 days of antibacterial treatment, and followed an uneventful course without recurrence. The GBS isolate was later identified as serotype Ia and multilocus sequence type (ST) 23. MICs against ampicillin, cefotaxime, and erythromycin were 0.06, 0.06, and $>1 \mu g/mL$, respectively. Rectovaginal and breast milk cultures of the mother during admission were negative.

**Discussion**

ASP is an uncommon disease usually seen in neonates or the elderly [1,2]. A review of 44 neonatal cases of ASP spanning four decades indicated that the major causative etiology was *S. aureus*, accounting for 61%, while GBS was isolated in only one case (2%) [3]. ASP caused by GBS was first reported in 1999 [7], and Walter et al. thereafter reviewed 5 such cases published between 1999 and 2009 [8]. For greater insight into this condition, we updated the review using Medline databases with key words of parotitis and GBS (or *Streptococcus agalactiae*). We found 11 relevant infants, including our case and 4 reports in non-English languages [4–11] (Table 1).

Several important findings were derived from this review. First is the infection route. Diagnosis of ASP is based on parotid swelling, purulent exudation from the Stensen’s duct, and the growth of pathogenic bacteria in culture of the pus [2]. Retrograde bacterial flow from the oral cavity into the parotid gland via the Stensen’s duct has been thought to be a major route of ASP. Indeed, according to a review by Ismail et al., of 30 neonates with ASP due to pathogens other than GBS in whom blood cultures were examined, only 10 were positive for blood cultures while all except two (one each for *Streptococcus pyogenes* and coagulase-negative *Staphylococcus*) were positive for pus cultures [3]. In contrast, all of the 11 GBS ASP infants had bacteremia, but purulent discharge from the Stensen’s duct was detected in only one case [4–11]. Thus, in comparison between infants with ASP due to GBS and non-GBS pathogens, the proportion of bacteremia (11/11 vs 10/30; p = 0.001, Fisher exact test) and positive pus discharge (1/11 vs 28/30; p < 0.001) had significant differences. These findings indicate that GBS invades the bloodstream and then spreads to the parotid gland in ASP infants. Routine laboratory data are nonspecific, and blood cultures are essential for diagnosis.

Second is age at onset. In the review of 11 infants, median age at onset was 30 days, ranging between 13 days and 12 weeks. Based on the time of onset, neonatal GBS is classified into early-onset disease (EOD) (0–6 days), late-onset disease (LOD) (7–89 days), and very-late onset disease (90 days and thereafter) [12,13]. Our review indicated that GBS ASP infants exclusively manifested as LOD. The clinical diagnoses of GBS LOD comprised of bacteremia without an infectious focus (65%), meningitis (25%), and cellulitis or osteoarthritis (2–3% each) [13]. Our case and the updated review added a rare infectious focus of ASP in GBS LOD.

Third is the presenting symptom. Five of the 11 GBS ASP infants appeared severely ill at initial evaluation, presenting with respiratory failure, septic shock, and/or hypoperfusion [4,7–10]. In our case, erythematous swelling over the parotid area rapidly extended to the submandibular region in 2–3 h. Nevertheless, the prognoses were good without any fatal outcome, probably because of early initiation of antimicrobial treatment. The symptoms differ from those in *S. aureus* ASP in that the affected patients are not associated with serious symptoms [3]. Such disparity might be explained by the fact that GBS ASP arises from systemic infection while *S. aureus* ASP is usually a focal infection. Two infants were born preterm; both appeared toxic and required mechanical ventilation at onset, and the remaining 9 had no predisposing conditions. As observed in all types of infant GBS disease [12,13], prematurity is a risk factor.

Fourth is the association with recurrent infections. Two of the 11 GBS ASP infants had had recurrent episodes of invasive GBS infection; one was as the first of two infectious episodes, and the other was the second of three. This recurrence rate is much higher than that estimated for GBS EOD and LOD, i.e., around 1–3% [13,14]. However, whether GBS ASP is more likely to be associated with recurrence requires further study, because of the limited number of reported cases.

Finally, we found that of the six isolates serotyped, serotype III (n = 4) was dominant, followed by Ia (n = 2). We furthermore, for the first time, examined multilocus ST of the isolate, showing as ST23. These findings of serotype distribution and the link between serotype Ia and ST23 were comparable with those observed for all GBS LOD [12,13,15].

In conclusion, this study demonstrates that GBS can cause ASP in a healthy newborn as LOD. While retrograde bacterial flow via

![Fig. 1. Coronal (A) and axial (B) images of contrast-enhanced computed tomography of the neck on admission, showing enlargement of the right parotid gland (*) with swelling of adjacent soft tissues (white arrow).](image-url)
| Case | Age/sex | Gestational age (weeks) | Birth weight (g) | Physical findings on admission | Laboratory data | Recurrent infectious episode | Prognosis | Ref |
|------|---------|-------------------------|------------------|-------------------------------|----------------|-----------------------------|-----------|-----|
|      |         |                         |                  |                               |                |                             |           |     |
| 1    | 35 day/F | 27                      | 959              | afebrile                      |                 |                             |           |     |
|      |         |                         |                  |                               | 27             | ND                          | Positive  | Ia  |
|      |         |                         |                  |                               | 27             | ND                          | Positive  | Ia  |
| 2    | 23 day/M | 41                      | 3200             | febrile                       |                 |                             |           |     |
|      |         |                         |                  |                               | 7.2            | 1.5                         | 29        | Positive III |
|      |         |                         |                  |                               | 7.2            | 1.5                         | 29        | Positive III |
| 3    | 53 day/F | Term                    | 2340             | febrile                       |                 |                             |           |     |
|      |         |                         |                  |                               | 15.2           | 8.8                         | ND        | Positive III |
|      |         |                         |                  |                               | 15.2           | 8.8                         | ND        | Positive III |
| 4    | 27 day/F | Term                    | ND               | febrile                       |                 |                             |           |     |
|      |         |                         |                  |                               | 10.4           | 13.3                        | 10        | Positive ND |
|      |         |                         |                  |                               | 10.4           | 13.3                        | 10        | Positive ND |
| 5    | 30 day/M | Term                    | ND               | febrile                       |                 |                             |           |     |
|      |         |                         |                  |                               | 2.13           | 17.7                        | 3         | Positive ND |
|      |         |                         |                  |                               | 2.13           | 17.7                        | 3         | Positive ND |
| 6    | 43 day/M | Term                    | ND               | febrile                       |                 |                             |           |     |
|      |         |                         |                  |                               | 9.9            | <1.0                        | ND        | Positive ND |
|      |         |                         |                  |                               | 9.9            | <1.0                        | ND        | Positive ND |
| 7    | 48 day/M | Term                    | ND               | septic shock                  |                 |                             |           |     |
|      |         |                         |                  |                               | 5.7            | <1.0                        | ND        | Positive III |
|      |         |                         |                  |                               | 5.7            | <1.0                        | ND        | Positive III |
| 8    | 12 week/M| 26                      | 915              | afebrile                      |                 |                             |           |     |
|      |         |                         |                  |                               | 23             | 7.6                         | ND        | Positive ND |
|      |         |                         |                  |                               | 23             | 7.6                         | ND        | Positive ND |
| 9    | 3 week/F | Term                    | ND               | septic shock                  |                 |                             |           |     |
|      |         |                         |                  |                               | 8.5            | 7.5                         | ND        | Positive III |
|      |         |                         |                  |                               | 8.5            | 7.5                         | ND        | Positive III |
| 10   | 13 day/M | 38                      | 3110             | febrile                       |                 |                             |           |     |
|      |         |                         |                  |                               | 14.2           | 2.25                        | 20        | Positive ND |
|      |         |                         |                  |                               | 14.2           | 2.25                        | 20        | Positive ND |
| 11   | 21 day/M | 38                      | 2933             | febrile                       |                 |                             |           |     |
|      |         |                         |                  |                               | 4.3            | 1.3                         | 15        | Positive Ia |
|      |         |                         |                  |                               | 4.3            | 1.3                         | 15        | Positive Ia |

ND, not documented; WBC, white blood cell; CRP, C-reactive protein; AMY, amylase; GBS, group B streptococcus; Ref, Reference.

* Age at onset of parotitis in cases associated with recurrent episodes.
the Stensen’s duct is a major infectious route in ASP due to non-GBS pathogens, GBS ASP primarily arises from bloodstream infection. Blood cultures are indispensable for accurate diagnosis and optimal therapy.

Informed consent

Written informed consent was obtained from the patient’s parents for the publication of this case report.

Authorship statement

All authors meet the ICMJE authorship criteria.

Ethical approval

Not required.

Funding source

None.

CRediT authorship contribution statement

Nagisa Ujita: Writing - original draft. Yu Kawasaki: Writing - original draft. Kousaku Matsubara: Writing - review & editing. Supervision. Kaya Kim: Writing - review & editing. Akiyoshi Naito: Writing - review & editing. Masayuki Hori: Writing - review & editing. Kenichi Isome: Writing - review & editing. Aya Iwata: Writing - review & editing. Yoshimichi Yamaguchi: Writing - review & editing. Bin Chang: Data curation, Writing - review & editing.

Declaration of Competing Interest

None to declare.

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