



## Case Report

# Fatal Mastitis due to Group a Streptococcus: An Autopsy Case

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### Abstract

**Background:** Severe infection by Streptococcus pyogenes, a type of group A Streptococcus (GAS), is uncommon but has a high fatality rate. We herein report the first autopsy case of mastitis due to GAS.

**Case presentation:** A healthy woman in her 40s died 3 days after complaining of severe right shoulder-to-chest pain. Forensic autopsy revealed a reddish-brown area with an irregular superficial abrasion extending from the neck to chest with thickened subcutaneous connective tissue. Microbiologically, the presence of GAS was identified from a blood culture collected at the time of autopsy. Histopathologically, numerous neutrophilic leukocyte and lymphocyte infiltrations were found around the mammary gland in the right breast. These changes were considered to indicate severe mastitis. The same infiltrations were observed in the liver and adrenal medulla. These findings were in accordance with systemic bacteraemia. Therefore, the patient was diagnosed with GAS-induced sepsis secondary to right mastitis. She was a non-lactating woman and had no lesions or risk factors related to GAS. This case illustrates that mastitis can lead to rapidly progressive GAS infection with a fatal outcome.

**Conclusions:** Early recognition of GAS infection and prompt initiation of supportive care with antibacterial therapy are required to prevent sudden death, even in patients with mastitis.

**Keywords:** Streptococcus Pyogenes; Group a Streptococcus; Fatal Mastitis; Bacteraemia due to GAS; Case Report

### Introduction

Streptococcus pyogenes, a type of group A Streptococcus (GAS), is a  $\beta$ -haemolytic gram-positive coccus that often causes upper respiratory tract inflammation and skin/soft tissue infection. GAS is a known colonizer of the oropharynx, genital mucosa, rectum, and skin, and its carriage rate in healthy adults is estimated at <5% in the throat and 1% in the vagina and rectum [1]. Despite the rarity of severe GAS infection, such as that causing bacteraemia, cellulitis, puerperal sepsis, meningitis, necrotizing fasciitis, and septic arthritis, such infections are serious and have a

high fatality rate. Invasive GAS disease is diagnosed when GAS is isolated from a normally sterile site, such as blood, subcutaneous tissues, surgical specimens, or bone [2]. According to previous studies, while the complication rate of invasive GAS disease shows remarkable consistency among industrialized nations (approximately 2 to 4 per 100,000 individuals), the fatality rate ranges from 8% to 19% [3,4]. The soft tissue is the most common site of invasive GAS infections, accounting for about half of all cases [3]. The most common skin and soft tissue infection sites are the extremities and perineum [4]. To the best of our knowledge, no reports have described fatal GAS infection of the soft tissue of the breast. We herein report a rare autopsy case of mastitis due to GAS.

## Case Presentation

### Overview

A woman in her 40s with no medical history except shoulder stiffness and low back pain developed right shoulder pain 3 days prior to her death. On the day of presentation, she was referred to an orthopaedist and was prescribed nonsteroidal anti-inflammatory drugs under the diagnosis of muscle strain. Soon after returning home, she fell asleep because of fatigue. The next day (2 days prior to her death), the pain had moved from her right shoulder to her right chest. Her husband confirmed that her right shoulder was red and swollen that night. The day before her death, she had developed severe shoulder to chest pain but went to work as usual. After returning home, she fell asleep with a cold pack on her chest because she was feverish with severe right shoulder-to-chest pain. At midnight, her husband heard her having a nightmare. At 6:00 AM the next morning, her husband found her dead on the bed. To determine the mechanisms and cause of death, a forensic autopsy was performed 27 hours after discovery of the body.

### Autopsy findings

The woman's height was 160.0 cm and weight was 56.2 kg. The surface of the body was light red, and purple-red post-mortem lucidity was observed on the back. No petechial haemorrhages were present on the oral mucosa or bilateral palpebral conjunctivae. A reddish brown area measuring  $28.5 \times 34.0$  cm with an irregular superficial abrasion extended from the neck to the right shoulder and to the right chest (Figure 1). No marked injuries were noted externally. Internally, no subcutaneous or intermuscular haemorrhage was observed in the chest. No marked abnormality was found in the shoulder joint. The subcutaneous region around the shoulder showed no abnormalities except for mild edema. From immediately below the right breast to the centre of the chest (Figure 2), the subcutaneous connective tissue was thickened to  $10.0 \times 10.0 \times 3.0$  cm. A small amount of dark red soft coagulated blood was observed in the heart cavities. No injuries or abnormalities with the exception of congestion were found in the cerebrospinal regions or in the intrathoracic and abdominal organs.

### Toxicological findings

No alcohol was detected in the cardiac blood or urine by gas chromatography. Urine drug screening tests with the INSTANT-VIEW® M-I immunoassay kit revealed no psychotropic or illicit drugs.

### Microbiological findings

A bacterial culture was performed using the blood collected at the time of autopsy, and GAS was identified. No other bacteria or fungi were detected.

### Histopathological findings

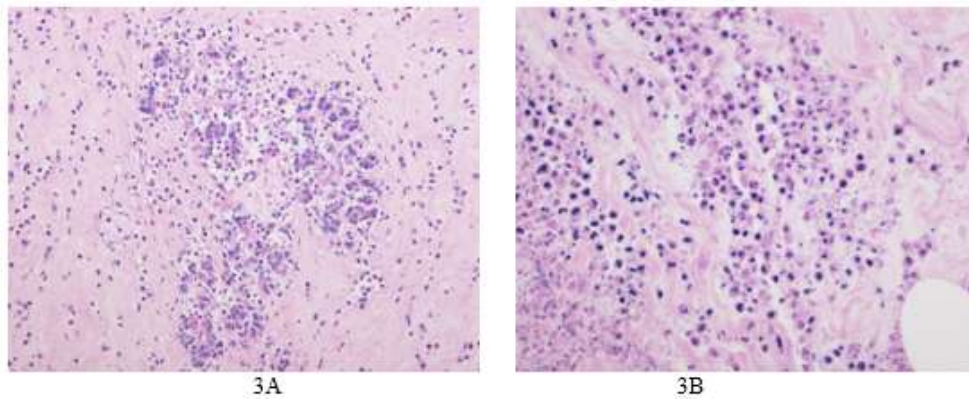
Numerous neutrophilic leukocyte and lymphocyte infiltrations were found around the mammary gland in the right breast (Figure 3). Immunohistochemical staining of the mammary gland tissue was performed to check for malignancy, and both p63 and CK5/6 were negative. These changes were considered to indicate severe mastitis. Marked neutrophilic leukocyte and lymphocyte infiltrations were also observed near Glisson's capsule in the liver (Figure 4), and lymphocyte infiltration was found in the adrenal medulla (Figure 5). These findings were consistent with systemic bacteraemia.



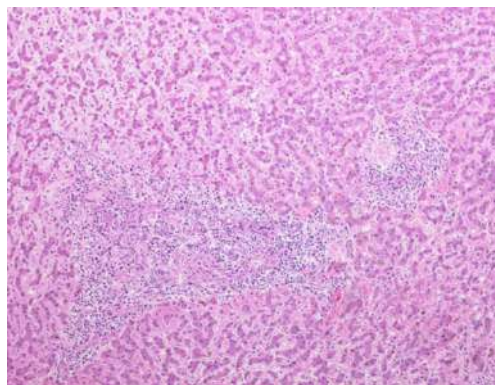
**Figure 1:** A reddish brown area with an irregular superficial abrasion extended from the neck to the right shoulder and to the right chest.



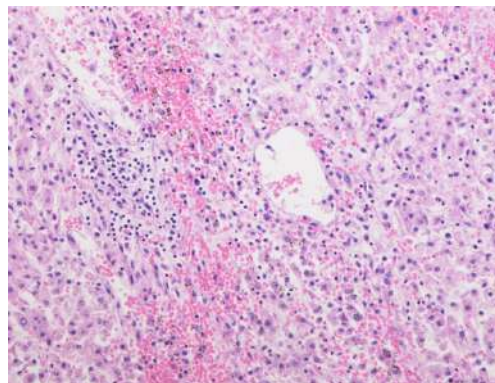
**Figure 2:** The subcutaneous connective tissue was thickened from immediately below the right breast to the center of the chest.



**Figure 3:** The mammary gland in the right breast. (A) Normal mammary gland structure and lymphocyte infiltrations were observed. (B) Bacterial colonies and numerous neutrophilic leukocyte and lymphocyte infiltrations were found (hematoxylin and eosin stain). Magnification,  $\times 400$ .



**Figure 4:** Glisson's capsule in the liver. Infiltrations of many neutrophilic leukocytes and lymphocytes were observed (hematoxylin and eosin stain). Magnification,  $\times 200$ .



**Figure 5:** Adrenal medulla. Lymphocyte infiltration was found (hematoxylin and eosin stain). Magnification,  $\times 400$ .



## Diagnosis of cause of death

Immediately below the right breast, subcutaneous connective tissue was thickened and histopathologically it was determined as severe mastitis. Furthermore, GAS was identified in the blood culture, and findings of sepsis were shown in other organs. Based on the autopsy, microbiological, and histopathological findings, the woman was diagnosed with GAS-induced sepsis secondary to right mastitis.

## Discussion

In this case, the patient died 3 days after developing right shoulder pain, highlighting the rapid progression of the disease. In a study by Lamagni et al. [5], invasive GAS diseases rapidly worsened, and 19% of the patients with this infection died within 7 days of diagnosis. Lepoutre et al. [6] reported that 68% of all deaths caused by invasive GAS diseases occurred within 4 days following the onset of infection. Therefore, some cases of GAS infection were recognized after sudden death, as in the present case, and forensic pathologists often diagnose GAS infection [7]. In this case, GAS was detected in the blood culture, and we considered that the patient had died of sepsis. In 82% of the skin and soft tissue infections caused by invasive GAS, the clinical condition progresses to bacteraemia [6]. Skin lesions (either traumatic, surgical, or due to a chronic skin condition), diabetes, an immunocompromised state, and injection drug use are risk factors for invasive GAS infection; however, no particular risk factor is identified in 21% to 23% of patients. In the present case, the patient was a non-lactating woman and had no lesions or risk factors related to GAS. Furthermore, the autopsy and histopathological findings revealed marked infectious change of the soft tissues compatible with mastitis. Therefore, the GAS infection was considered to have originated from right mastitis. Only one other case of GAS due to mastitis has been reported to date, and the patient fully recovered [8]. Therefore, this is the first case of fatal GAS infection due to mastitis.

## Conclusions

We experienced a rare autopsy case of mastitis due to GAS. Prompt intervention is important to save the lives of patients with this infection.

## Disclosure

Author Contributions: MH made substantial contributions to the conception of the work. AT analysed and interpreted the autopsy data regarding the mastitis due to group A Streptococcus, and drafted the original manuscript. KM performed the histological examination of the mammary gland, and was a major contributor in writing the manuscript. MF, MT and MN substantially contributed to the revision of the manuscript drafts. All authors read and approved the final manuscript.

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**Informed Consent Statement:** Written informed consent was obtained from the next of kin (9 August 2016).

**Data Availability Statement:** Not applicable.

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**Conflicts of Interest:** None.

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