

# The co-existence of Lemierre's syndrome and Bezold's abscesses due to *Streptococcus constellatus*

## A case report

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### Abstract

**Rationale:** The ancient infectious diseases, Lemierre's Syndrome and Bezold's Abscesses are rare.

**Patient concerns:** A 70-year-old Japanese woman with a 15-year history of Parkinson's disease was referred to our hospital due to fever, occipital headache and bilateral shoulder pain that had continued for three months. She had been prescribed prednisolone due to a diagnosis of polymyalgia rheumatica.

**Diagnoses:** A blood culture revealed bacteremia of *Streptococcus constellatus*. In addition, computed tomography revealed Bezold's abscesses and Lemierre's syndrome.

**Interventions:** We administered ceftriaxone for 31 days, followed by oral amoxicillin.

**Outcomes:** The patient recovered and the abscesses improved.

**Lessons:** This case underscores the importance of blood culture tests and cross-referencing with radiological imagings in the diagnoses of these rare critical infectious diseases that mimic polymyalgia rheumatica.

**Abbreviation:** PMR = polymyalgia rheumatica.

**Keywords:** Bezold's abscess, Lemierre's syndrome, polymyalgia rheumatica, *Streptococcus constellatus*

## 1. Introduction

The first report of Bezold's abscesses and Lemierre's syndrome were noted in ancient times (1881 and 1936), and both continue to be regarded as rare and critical diseases.<sup>[1,2]</sup> For these and other examples, rarity often results in delayed diagnoses. In a review of 17 cases of Bezold's abscesses, the needed duration of diagnosis was 2 days to 2 weeks.<sup>[3]</sup> In a similar manner, a review of 14 cases of Lemierre's syndrome revealed that the amount of time needed for a correct diagnosis ranged from 2 to 23 days.<sup>[4]</sup> In

Japan, diagnostic imagings that include computed tomography have been treated casually in secondary or tertiary care hospitals. And in primary care settings, these forms of diagnosis were often overlooked due to low levels of accessibility to imaging modalities. Herein, we report a case involving the co-existence of Bezold's abscesses and Lemierre's syndrome that was previously misdiagnosed as polymyalgia rheumatica (PMR). We present this case and a review of the literature to discuss the pathophysiology of the co-existence of 2 rare infectious diseases that underscores the critical importance of recognizing PMR-mimicking symptoms.

## 2. Case report

A 70-year-old woman was referred to our hospital due to fever, occipital headache and bilateral shoulder pain that had continued for 3 months. One month prior to the referral, her physician had prescribed oral prednisolone 20mg per day based on the diagnosis of PMR. The regimen of prednisolone was immediately discontinued because it worsened her symptoms of Parkinson's disease. However, prednisolone was restarted due to a sustained fever (38°C) and musculoskeletal symptoms. The dosage of the prednisolone was raised to 30mg a day 2 weeks before the referral. The patient had been admitted to another hospital due to high-grade fever (40°C) 3 days before the referral. A blood culture tested positive for *Streptococcus constellatus* and a regimen of intravenous ampicillin/sulbactam was started. For further investigation and treatment, the patient was referred to our hospital. The patient had a medical history of Parkinson's disease treated for 15 years.

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Ethics: Written informed consent for the publication of this case report was obtained from the patient. Also Kurume University Hospital Ethics Committee approved the submission.

The authors have no conflicts of interest to disclose.

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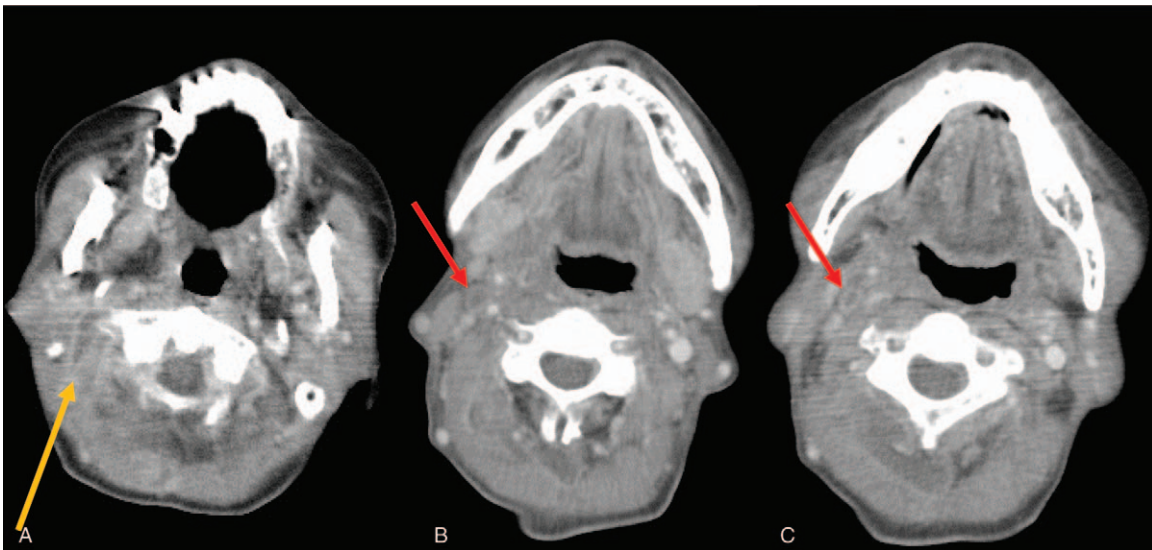
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**Figure 1.** Enhanced computed tomography revealed multiple abscesses on the neck (yellow arrow: Bezold's abscess) (A) and thrombosis in the right internal jugular vein (red arrow) (B, C).

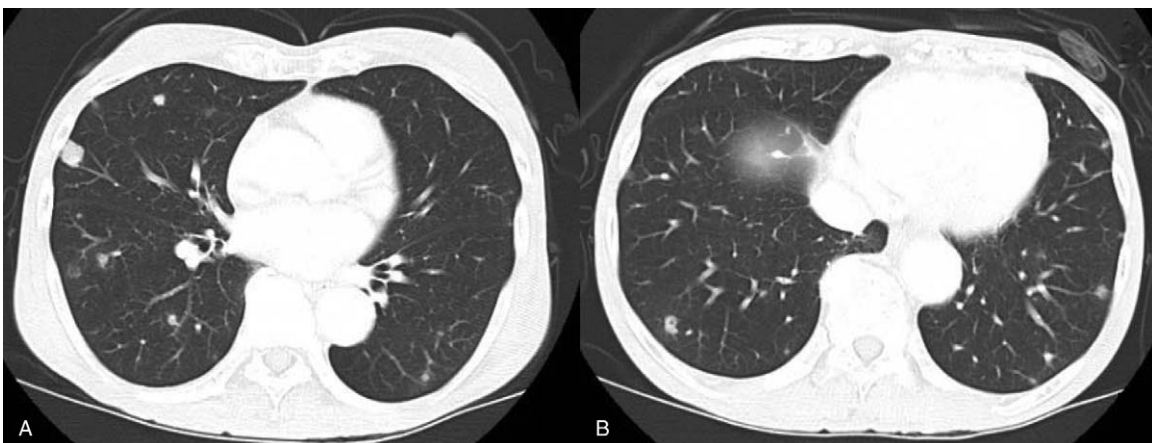
At the time of admission, the patient's vital signs were as follows: blood pressure, 88/57 mmHg; heart rate, 65 beats/min; respiratory rate, 18 breaths/min; body temperature, 37.7°C and Glasgow Coma Scale, E4V5M6 (total 15/15). Her height was 156.5 cm and her body weight was 41.3 kg. Upon physical examination, tenderness was noted in her neck. Neither arthritis nor emboli on extremities were detected on her extremities. The patient's oral hygiene was checked by dentists and showed no caries. In addition, otolaryngologists detected right otitis media.

Laboratory data revealed low levels of plasma protein (total protein: 5.65 g/dL, albumin: 2.32 g/dL) and high levels of inflammation (c-reactive protein: 11.03 mg/dL, erythrocyte sedimentation rate: 75.0 mm/h, white blood cell count: 10,600/ $\mu$ L (neutrophil: 96.1%, lymphocyte: 2.4%)). Contrast-enhanced computed tomography showed polycystic lesions (abscesses) on her right posterior and on the lateral region of her neck, thrombosis in her right internal jugular vein (Fig. 1A–C) and multiple nodules on her bilateral lung fields (septic emboli) (Fig. 2A and B). Magnetic resonance imaging confirmed the polycystic lesions of her neck, and thrombosis in her right

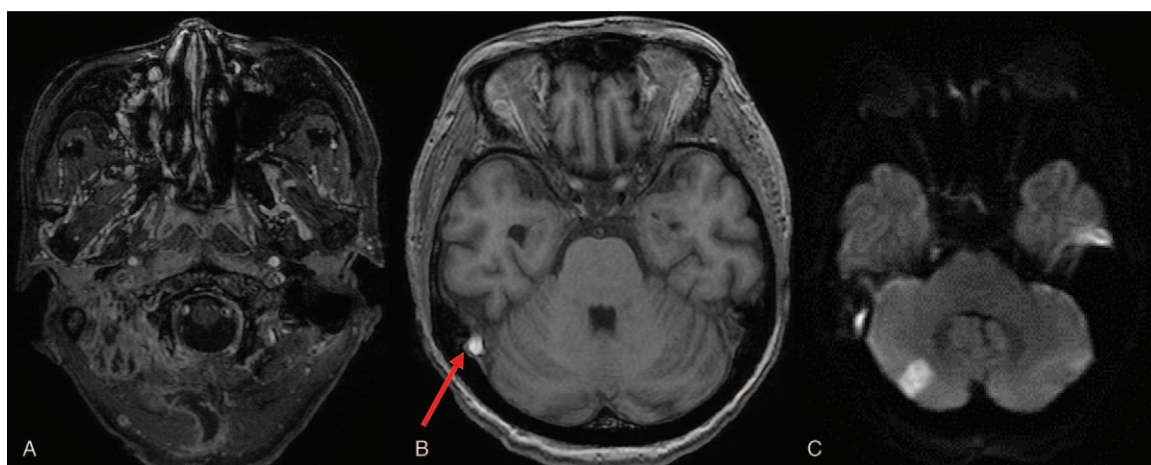
transverse sinus (Fig. 3A and B). Diffusion weighted magnetic resonance imaging also revealed acute infarction of the right cerebellum (Fig. 3C). Transthoracic echocardiography was also performed, and neither infective endocarditis nor severe regurgitation was observed.

Based on her symptoms and the results of imaging and microbiological tests, we concluded that disseminated *S. constellatus* infection with Bezold's abscesses and Lemierre's syndrome was the definite diagnosis. We suspected the first lesion was right otitis media followed by Bezold's abscesses, which caused PMR-like symptoms (fever and musculoskeletal symptoms). Prednisolone therapy leads to the hematogenous dissemination of bacteria that forms Lemierre's syndrome.

We started ceftriaxone at a dose of 2 g (the susceptibility to penicillin was not obtained on the day of admission) administered intravenously every 24 hours after drawing a blood culture. However, the blood cultures on the day of admission were already negative for bacteria. The fever and musculoskeletal symptoms soon subsided, and computed tomography was performed again to evaluate abscesses and septic emboli on



**Figure 2.** Computed tomography showed multiple nodules (septic emboli) on bilateral lung fields (A, B).



**Figure 3.** Cervical abscesses were also confirmed by enhanced magnetic resonance imagings (A). The thrombosis on the right transverse sinus was detected on T1-weighted magnetic resonance imagings (red arrow) (B). Diffusion weighted magnetic resonance imagings confirmed the acute infarction of the right cerebellum (C).

the lungs, which had declined without drainage. The prednisolone regimen was tapered gradually, and the PMR-like symptoms did not reoccur. We assumed the PMR-like symptoms were caused by the bacteremia. In addition, anticoagulants (heparin, followed by warfarin) were used because of the large thrombosis in not only the internal jugular vein, but also in the transverse sinus.<sup>[5]</sup>

At 33rd hospital day, the patient was discharged with a prescription for 1500 mg per day of amoxicillin taken orally for the remaining abscesses on the neck and for the septic emboli on the lungs. The susceptibility to ampicillin had been obtained from another hospital, and the minimum inhibitory concentration was 0.12  $\mu\text{g}/\text{mL}$  (susceptible). We continued the oral amoxicillin, warfarin and a low dose of prednisolone for half a year after the patient's discharge. We used enhanced computed tomography to document the sufficient decline of the abscesses, before stopping all medication. No recurrence of infection or PMR-like symptoms was seen 3 months after the end of the amoxicillin and prednisolone regimens.

### 3. Discussion

Our case was a Japanese woman with the co-existence of Bezold's abscesses and Lemierre's syndrome in the modern era. The relationship of 2 different diseases are poorly understood, although Bezold's abscess could cause Lemierre's syndrome.<sup>[6]</sup> We searched PubMed, Google Scholar, and Ichushi (the Japanese database for medical literature and conference proceedings) databases for similar case reports written in either English or Japanese, and found several cases of co-existence had been reported.<sup>[7,8]</sup> In a recent report,<sup>[8]</sup> the first infection was otitis media, as with our case, but no risk factors were documented for complications that could lead to Bezold's abscesses and Lemierre's syndrome.

Lemierre's syndrome normally is followed either by tonsillitis or by an upper respiratory tract infection.<sup>[2]</sup> The first symptom commonly is a sore throat.<sup>[2]</sup> In our case, dentists performed an oral hygiene examination during treatment and found no abnormalities. Left otitis media was observed, and sore throat did not precede the bacteremia. These clinical findings suggested that otitis media first spread to Bezold's abscesses, and

improvident steroid therapy for musculoskeletal symptoms had likely caused the dissemination of *S. constellatus*. This organism is compatible with the pathogen of Bezold's abscesses.<sup>[9]</sup> Finally, it is possible that the bacteremia and direct influence of cervical inflammation had caused Lemierre's syndrome.

At first, our patient's primary care physician had initially suspected that PMR was causing the musculoskeletal symptoms that were subsequently diagnosed as being caused by cervical abscesses and bacteremia. A variety of condition is known to mimic PMR or cause polymyalgia symptoms.<sup>[10]</sup> Atypical features that include sustained high fever and a lack of response to steroids should indicate the presence of a condition (eg. infection, vasculitis) other than isolated PMR. Specific clinical symptoms and serum biomarker are not identified for a diagnosis of PMR, which means an exclusion diagnosis for infectious diseases and malignancy is indispensable. Neck tenderness was significant with our patient and the surface of her skin was not reddish. This complicated a correct diagnosis without imagings and a blood culture. Finally, after stopping corticosteroid therapy, the musculoskeletal symptoms did not reoccur. These findings and the clinical course revealed that the patient's PMR was a case of misdiagnosis.

The patient's clinical course after confirming the diagnosis was favorable; however, Lemierre's syndrome is often fatal even in a recent review.<sup>[11]</sup> The mortality of Lemierre's symptoms was reported as 0% to 18%.<sup>[11]</sup> In our case, fortunately, the blood culture promptly became negative. In addition, a long duration of treatment with close follow-up by contrast-enhanced computed tomography might effectively improve a patient's outcome.

### 4. Conclusion

This patient experienced the co-existence of Bezold's abscesses and Lemierre's syndrome. The patient's initial diagnosis was PMR; however, blood culture and imagings led to the true diagnosis. Fortunately, the patient recovered with no sequelae. Following the treatment for infectious diseases, the PMR-like symptoms did not reoccur. Clinicians should be careful to rule out critical diagnoses, particularly those of infectious disease, that mimic PMR.

## Author contributions

**Supervision:** Shinjiro Kaieda, Hiroaki Ida, Hiroshi Watanabe.

**Writing – original draft:** Kenichiro Yaita.

**Writing – review & editing:** Suzuna Sugi, Makiko Hayashi, Takuma Koga, Tomohiro Ebata, Yoshiro Sakai.

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