

CASE REPORT

Postmortem diagnosis of gestational psittacosis: A case report

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Abstract

Background: Gestational psittacosis is one of the causes of unanticipated maternal death but has been difficult to diagnose early in clinical practice.**Case Presentation:** A 28-year-old woman who was 7 months pregnant experienced flu-like symptoms, which deteriorated. She was brought to our hospital in shock, and the fetus was nonviable. The patient was diagnosed with pneumonia and septic shock and administered meropenem. Despite aggressive resuscitation, she died 7h after symptom onset. After obtaining consent from the patient's family, the autopsy was done to identify the cause of death. Microscopically, there was intervillous neutrophil accumulation in the placenta. Genetic analysis detected the *Chlamydia psittaci* gene in several organs, including placenta.**Conclusion:** Gestational psittacosis should be considered for a pregnant woman with flu-like symptoms. Moreover, unanticipated death of a pregnant woman might warrant a detailed autopsy to reveal the cause of death.

KEY WORDS

autopsy, chlamydia, gestational psittacosis, pregnant, shock

INTRODUCTION

Psittacosis is generally known as a mild pulmonary infection caused by *Chlamydia psittaci*. Occasionally, in pregnant women, it can rapidly worsen and lead to fatal outcomes.¹⁻⁷ Because of difficulties in early diagnosis, some cases of maternal death of unknown cause may be secondary to undetected gestational psittacosis. The patient in this report had a worsening and fatal clinical course and was diagnosed with gestational psittacosis only on autopsy. This case emphasized the importance of early diagnosis and postmortem autopsy.

CASE

A 28-year-old woman who was 7 months pregnant experienced persistent fever and myalgia. Both tests for severe acute respiratory syndrome coronavirus 2 (SARS-Cov-2) antigen and influenza were negative. The patient initially

stayed at home and took acetaminophen but eventually felt worse, with fever, confusion, and restlessness. Thereafter, she was immediately transported to our hospital.

On physical examination, the initial Glasgow Coma Scale was 11 (E4V2M5), heart rate was 150/min, blood pressure was 95/77 mmHg, respiratory rate was 40/min, and axillary temperature was 38.5°C. There were bilateral coarse crackles on auscultation. The blood test showed hypoxemia, lactic acidosis, thrombocytopenia, systemic inflammation, and coagulation dysfunction (Table 1). Both nasopharyngeal swab SARS-Cov-2 PCR and vaginal discharge antigen test for group A β -hemolytic streptococcus were negative.

She was immediately intubated and placed on mechanical ventilation. After massive fluid infusion and continuous noradrenalin administration at 0.1–0.5 μ g/kg/min, the systolic blood pressure reached approximately 100 mmHg. Echocardiography showed a hyperdynamic state but no right heart failure. Computed tomography showed bilateral nodular opacities with infiltration, including air bronchograms

This article is based on the study of "Postmortem diagnosis of gestational psittacosis: a case report" that was first reported by Nihon Kyukyu Igakukai Zasshi in the *Journal of Japanese Association for Acute Medicine*.

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TABLE 1 Biochemical analyses.

Blood gas analysis (room air)				
pH	7.14		LDH	1706 U/L
PCO ₂	21.9 mmHg		CK	187 U/L
PO ₂	63 mmHg		Na	127 mEq/L
lactate	8.7 mmol/L		K	4 mEq/L
			Cl	101 mEq/L
WBC	8990 ×10 ² /μL		BUN	18.1 mg/dL
Hb	16.6 g/dL		Cr	2.04 mg/dL
Ht	48.2 %		PCT	22.4 ng/mL
Plt.	0.9 ×10 ⁴ /μL		PT	30.3 %
			PT-INR	1.88
CRP	30.4 mg/dL		APTT	77.9 Sec
T-Bil	5.41 mg/dL		D-Dimer	300 μg/mL
AST	297 U/L		SARS-CoV-2	(-)
ALT	46 U/L		GAS antigen	(-)

Abbreviations: ALT, alanine aminotransferase; APTT, activated partial thromboplastin time; AST, aspartate aminotransferase; BUN, blood urea nitrogen; CK, creatine kinase; Cr, creatinine; CRP, C-reactive protein; GAS, Group A Streptococcus; Hb, hemoglobin; Ht, hematocrit; LDH, lactate dehydrogenase; PCT, procalcitonin; Plt., platelet; PT, prothrombin time; PT-INR, prothrombin time-international normalized ratio; SARS-CoV-2, severe acute respiratory syndrome coronavirus 2; T-Bil, total bilirubin; WBC, white blood cell.

in the dorsal pulmonary area (Figure 1A–C), the fetus in the uterus, and no other abnormalities in the abdominal and pelvic areas (Figure 1D, E). Ultrasound confirmed intrauterine fetal death, based on the absence of fetal heartbeat. After giving meropenem for severe pneumonia, the patient was admitted to the intensive care unit (ICU).

At the ICU, her blood pressure decreased again, and massive blood transfusion was administered. Despite aggressive fluid resuscitation and subsequent addition of vasopressin and catecholamines, the circulatory and respiratory status collapsed, and the patient finally succumbed to pulseless electrical activity 4 h after ICU admission. Despite cardio-pulmonary resuscitation, the patient died 7 h after the symptom onset. Although we proposed doing an autopsy to the family, they were initially extremely confused to accept it. We took time to explain the need to identify the cause of death, and they finally understood and consented.

On autopsy, there were minimal bleeding, congestion, and edema on the surface of skin, lung, liver, and spleen. On microscopic examination (Figure 2), there were no infectious changes in the lungs, but there were hemophagocytosis in the bone marrow, vascular obstruction in the nephron, and intervillous neutrophil accumulation in the placenta. The tissues were sent to Osaka Women's and Children's Hospital,

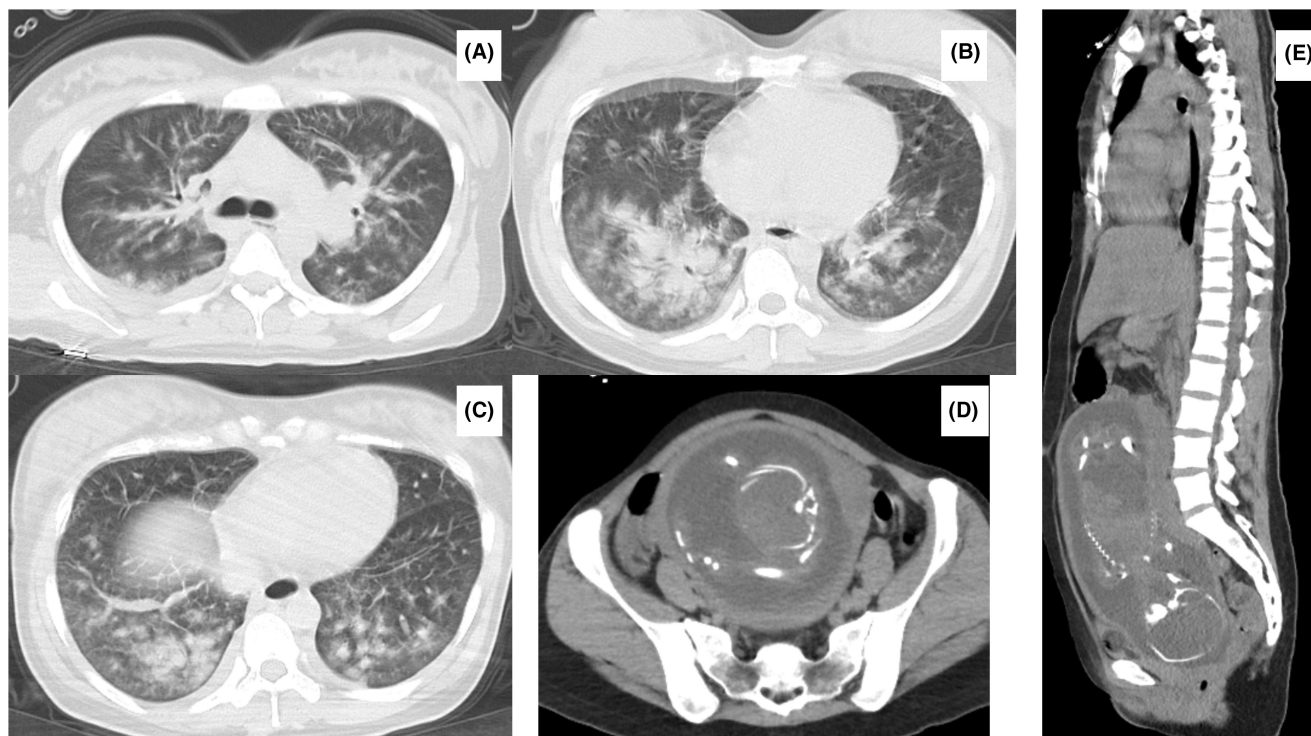


FIGURE 1 Computed tomography (CT) images on the day of hospital admission. (A–C) CT scan of the lungs shows bilateral consolidation with air bronchogram and infiltration, predominantly in the posterior area. (D, E) Abdominal CT scan shows no ascites, no abnormal inflammation, and the fetus in the uterus.

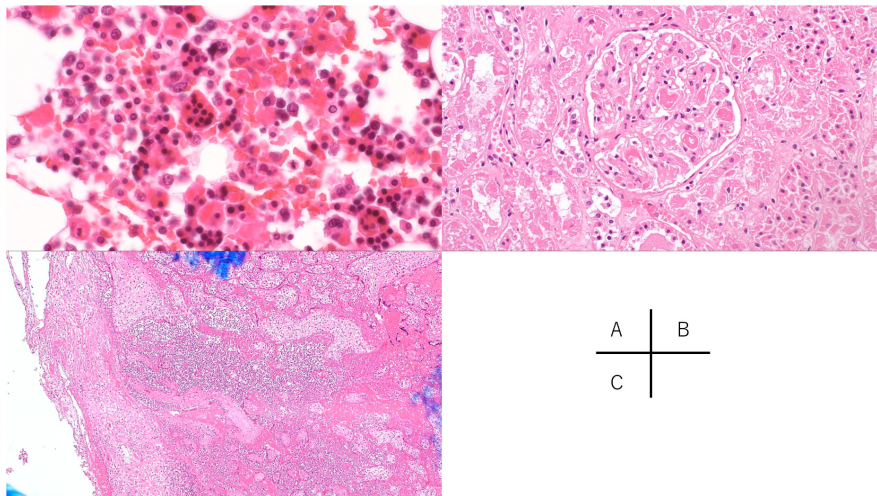


FIGURE 2 Histopathological findings. (A) There is hemophagocytosis in the bone marrow. (B) The glomerulus is damaged by a thrombus occlusion. (C) In the placenta, there is intervillous neutrophil accumulation, which is the hallmark of gestational psittacosis (hematoxylin and eosin [H&E] stain).

and genetic examination proved the presence of *C. psittaci* in the placenta, lungs, liver, and spleen. Consequently, gestational psittacosis was diagnosed.

DISCUSSION

Although this case had an unfortunate demise, autopsy was able to reveal the final diagnosis of gestational psittacosis. In cases of unanticipated maternal death, efforts should be made to pursue the cause of death by autopsy. In 2010, the Japan Association of Obstetricians and Gynecologists started implementing a registration program that reviews all cases of maternal death in Japan.⁸ Despite the advocacy on its significance in identifying the cause of death, autopsy remains to have a rate of approximately 30% in Japan. Some of the factors for this low autopsy rate include limited public awareness, negative public connotation, and even incorrect understanding among physicians. However, sharing information obtained from autopsies may help decrease the rate of future maternal death. In this case, the family eventually understood the importance of identifying the cause of death and wished that their experience would help patients in the future.

Gestational psittacosis is a zoonotic disease that is caused by *C. psittaci*. Because of an intolerance for cellular immunity, maternal patients are susceptible to infection with intracellular parasitic microbe. Although early diagnosis is important, the serological test for *C. psittaci* takes time and has low sensitivity. To ensure a correct diagnosis, careful interview about occupations, a history of animal contact, and symptoms is essential. In this case, we could not obtain enough information from the patient on admission. After she died, the family told us that she was a nurse and had no recent animal contact. However, the laboratory data of hyponatremia and liver, renal, and cardiac dysfunction presented in this case could have been

indicators of *C. psittaci* infection.⁹ Air-bronchograms in the bilateral consolidations in the lung CT scan observed in this case were also common findings of *C. psittaci* pneumonia.⁹ Considering these findings comprehensively, we could have had a chance to suspect gestational psittacosis in this case. Additionally, the use of appropriate antibiotics, such as tetracyclines, could have ensured a better outcome for the patient.

In this case, we were simultaneously considering some differential diagnoses. The symptoms of hypoxia, shock, and coagulation dysfunction were consistent with amniotic embolism. Pulmonary embolism may sometimes cause acute respiratory failure in pregnant women. Fulminant hemolytic streptococcal infection causes progressive shock. These differential diagnoses were ruled out not during the clinical course but on autopsy.

To save critically ill pregnant patients, early fetus delivery following pregnancy termination or perimortem cesarean section is one of the options that can effectively release inferior vena cava compression and increase circulatory volume.¹⁰ In previous reports, some patients who underwent early fetus delivery survived gestational psittacosis.^{1,2,4} However, it was challenging in this case because of uncontrollable bleeding.

No infection was detected in the lungs on microscopic examination. However, the presence of the *C. psittaci* gene in several organs was proven by genetic examination, similar to a previously reported case of gestational psittacosis.⁵ As *C. psittaci* has a predilection for the placenta, the inflammation induced by *C. psittaci* hematogenously moved from the lungs to the placenta, thus leading to villitis. The subsequent impairment of fetoplacental blood flow and bacterial embolism caused placental dysfunction and fetal death, which provoked septic shock, abnormal blood distribution, and coagulation dysfunction in the mother. Consequently, multiple organ failure may have caused maternal death.

CONCLUSION

Gestational psittacosis should be kept in mind when managing pregnant patients with flu-like symptoms. In cases of unanticipated maternal death, autopsy should be employed to pursue the cause of death.

ACKNOWLEDGMENTS

We thank all medical staff members at Iwakuni Clinical Center for their help in treating this patient.

CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest for this article.

DATA AVAILABILITY STATEMENT

Data sharing is not applicable to this article as no new data were created or analyzed in this study.

ETHICS STATEMENT

The protocol for this case report has been approved by a suitably constituted Ethics Committee of the Iwakuni Clinical Center, Approval No. 0505, and it confirms to the provisions of the Declaration of Helsinki. All informed consent was obtained from the guardians.

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How to cite this article: Miyauchi T, Hirata Y, Fukuda S. Postmortem diagnosis of gestational psittacosis: A case report. *Acute Med Surg*. 2024;11:e932. <https://doi.org/10.1002/ams2.932>