

A Case of Legionella Disease Mimicking Dermatomyositis

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Abstract

Legionella disease is a severe form of pneumonia that can present with rare extrapulmonary manifestations, potentially leading to diagnostic confusion. We report a 61-year-old man with alcohol dependence and heavy smoking who presented with severe pneumonia and systemic erythema, including seborrheic dermatitis-like erythema on the face, erythematous rashes spreading across the trunk, and livedo reticularis on the lower limbs. Laboratory findings showed markedly elevated ferritin, and computed tomography revealed widespread infiltrates and ground-glass opacities. Although empiric antibiotics were initiated, the presence of atypical cutaneous findings and progressive pulmonary lesions raised suspicion for autoimmune-associated organizing pneumonia, and prednisolone was initiated. The nonspecific cutaneous manifestations raised suspicion for dermatomyositis, but no pathognomonic rashes, such as Gottron papules or heliotrope rash, were observed, prompting further investigation for infection before escalating immunosuppression. A urinary antigen test subsequently confirmed *Legionella pneumophila*. The patient recovered fully after treatment with levofloxacin. This case highlights that Legionella disease can present with pulmonary and nonspecific cutaneous manifestations that may raise suspicion for dermatomyositis. Clinicians should consider Legionella infection in patients with severe pneumonia and atypical extrapulmonary manifestations, particularly those with relevant risk factors.

Categories: Rheumatology, Internal Medicine, Infectious Disease

Keywords: dermaomyositis, diffuse rash, legionella disease, mimick, pulmonary, pulmonary disease

Introduction

Legionella disease is a potentially severe form of atypical pneumonia caused predominantly by *Legionella pneumophila*. In addition to pulmonary involvement, Legionella infection may cause extrapulmonary manifestations, including gastrointestinal, neurological, and hepatic findings [1]. Although cutaneous manifestations are rare, erythematous, maculopapular, and other nonspecific skin eruptions have been reported in patients with Legionella infection [2]. Thus, the combination of nonspecific skin eruptions and severe pulmonary disease may complicate the diagnosis.

Dermatomyositis is an idiopathic inflammatory myopathy characterized by cutaneous manifestations and inflammatory muscle involvement and is frequently associated with interstitial lung disease [3]. The cutaneous manifestations of dermatomyositis are diverse, ranging from pathognomonic skin rashes, such as Gottron papules and heliotrope rash, to nonspecific and rare cutaneous manifestations. Pulmonary involvement in dermatomyositis may be severe and potentially fatal, particularly in patients with rapidly progressive interstitial lung disease. In some patients, muscle involvement may be absent or clinically subtle, as in clinically amyopathic dermatomyositis. Therefore, dermatomyositis should not be overlooked when cutaneous manifestations are accompanied by severe pulmonary involvement.

Legionella infection accompanied by skin eruptions may complicate the differential diagnosis of dermatomyositis, although such cutaneous manifestations are uncommon and not well recognized. This distinction is clinically important because inappropriate immunosuppressive therapy may exacerbate the underlying infection.

This case highlights the diagnostic challenge posed by atypical cutaneous manifestations of Legionella infection and emphasizes the importance of carefully evaluating the morphology and distribution of skin lesions, including the presence or absence of pathognomonic manifestations of dermatomyositis, when assessing patients with severe pneumonia and extrapulmonary findings.

Case Presentation

A 61-year-old male with a long-standing history of alcohol dependence was referred to our hospital for treatment of worsening pneumonia. He had participated in an alcohol day-care program, and he had undergone detoxification in a psychiatric hospital whenever he experienced episodes of continuous drinking. He smoked 20 cigarettes daily (35 pack-years). On the tenth day of the sixth voluntary

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hospitalization at the previous hospital, the patient developed fever and right lower leg swelling. He was diagnosed with cellulitis and treated with piperacillin; however, the fever persisted. On the twelfth day of hospitalization, his respiratory status deteriorated, and chest radiographs revealed findings consistent with left-sided pneumonia. The antibiotic regimen was switched to ceftriaxone; however, his respiratory condition continued to decline. On the fifteenth day, after failing to improve with initial treatments, the patient was diagnosed with refractory pneumonia. Given the severity of his condition, he was transferred to our hospital for more intensive care and treatment. Upon transfer, the patient presented with disorientation, a fever of 39.4 °C, respiratory rate of 26 breaths per minute, blood pressure 136/90 mmHg, heart rate 109 bpm, and SpO₂ of 97% on nasal high-flow oxygen (70% at 60 L). Clinical examination revealed bilateral crackles in the bilateral lung fields, seborrheic dermatitis-like erythema on the face (Figure 1A), erythematous rashes spreading across the trunk (Figure 1B), and livedo reticularis on the lower limbs. The history regarding the progression of the rash could not be confirmed due to his confusion.

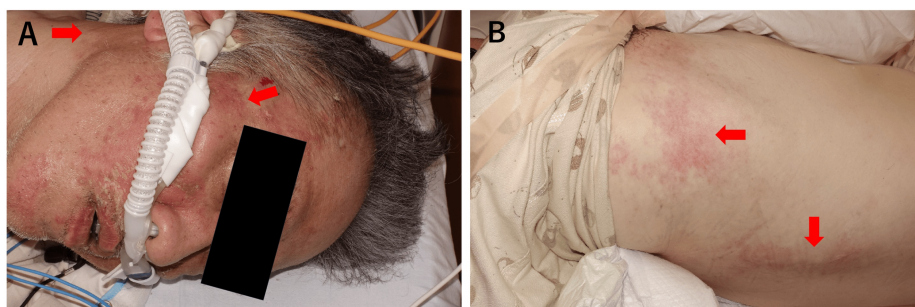


FIGURE 1: Images of skin findings

(A) Worsening facial erythema on a background of seborrheic dermatitis and faint erythema around the posterior neck and bilateral shoulders. (B) Map-like erythema over the back.

Blood tests showed serum sodium 129 mEq/L, aspartate aminotransferase 562 U/L, alanine aminotransferase 170 U/L, lactate dehydrogenase 594 U/L, creatine kinase 236 U/L, and markedly elevated ferritin at 15,600 ng/mL (Table 1).

Laboratory test	Result	Reference range
WBC count	15.64×10 ⁹ /L	3.30-8.60×10 ⁹ /L
RBC count	3.66×10 ⁶ /μL	4.35-5.55×10 ⁶ /μL
Hemoglobin	13.6 g/dL	13.7-16.8 g/dL
Platelet count	26.8×10 ⁴ /μL	15.8-34.8×10 ⁴ /μL
AST	562 U/L	13-30 U/L
ALT	170 U/L	10-42 U/L
LDH	594 U/L	124-222 U/L
CK	236 U/L	59-248 U/L
Total bilirubin	1.32 mg/dL	0.40-1.50 mg/dL
Albumin	2.5 g/dL	4.1-5.1 g/dL
BUN	12.6 mg/dL	8.0-20.0 mg/dL
Creatinine	0.79 mg/dL	0.65-1.07 mg/dL
Na	129 mEq/L	138-145 mEq/L
K	3.3 mEq/L	3.6-4.8 mEq/L
Cl	91 mEq/L	101-108 mEq/L
CRP	40.2 mg/dL	0.00-0.14 mg/dL
Ferritin	15603 μg/mL	39.4-340.0 μg/mL

TABLE 1: Baseline laboratory findings

WBC: white blood cell, RBC: red blood cell, AST: aspartate transaminase, ALT: alanine aminotransferase, LDH: lactate dehydrogenase, CK: creatine kinase, BUN: blood urea nitrogen, CRP: C-reactive protein

Chest radiography performed at our hospital showed bilateral pulmonary infiltrates with left pleural effusion (Figure 2A). Chest computed tomography (CT) revealed emphysematous changes in both lungs and diffuse, non-segmental ground-glass opacities throughout both lung fields. The margins of the ground-glass opacities were accompanied by areas of consolidation in both lower lobes. A small left-sided pleural effusion was also observed (Figure 2B). CT imaging was performed using an IQon Spectral CT scanner (Philips Healthcare, Tokyo, Japan).

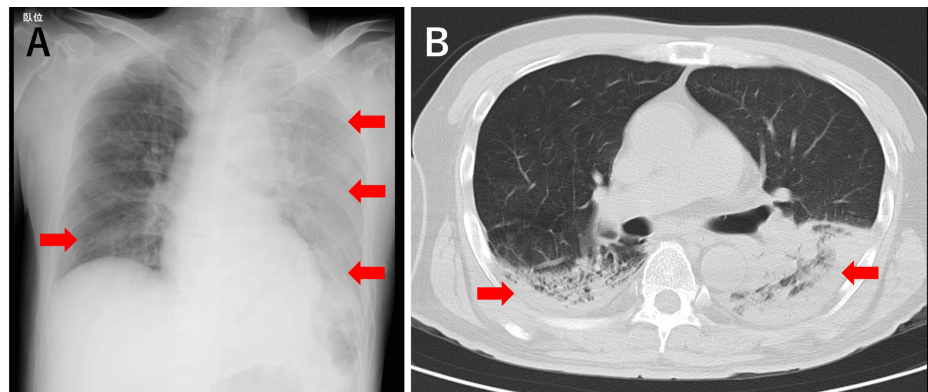


FIGURE 2: Chest X-rays and CT imaging

(A) Bilateral pulmonary infiltrates with left pleural effusion on chest radiography. (B) Areas of consolidation along the margins of the ground-glass opacities in both lower lobes and a small left-sided pleural effusion on chest CT. Chest CT was performed using an IQon Spectral CT scanner (Philips Healthcare, Tokyo, Japan).

Empiric therapy with cefepime and vancomycin was started for severe hospital-acquired pneumonia. Given the cutaneous findings and rapidly progressive pulmonary lesions, autoimmune diseases, including dermatomyositis, were included in the differential diagnosis. Because the pulmonary lesions were rapidly progressing and further deterioration was a concern, prednisolone was initiated on hospital day 2 while the diagnostic evaluation was ongoing.

Sputum culture obtained at our hospital yielded only oral flora, sputum culture for acid-fast bacilli was negative, and blood cultures obtained at our hospital were negative. Antinuclear antibodies and other dermatomyositis-specific or related antibodies were negative.

A biopsy of the erythematous lesion on the trunk was performed before initiation of prednisolone. Histopathological examination of the skin biopsy specimen showed a superficial perivascular inflammatory cell infiltrate in the dermis, predominantly composed of lymphocytes, on hematoxylin and eosin staining (Figures 3A, 3B). These findings were nonspecific.

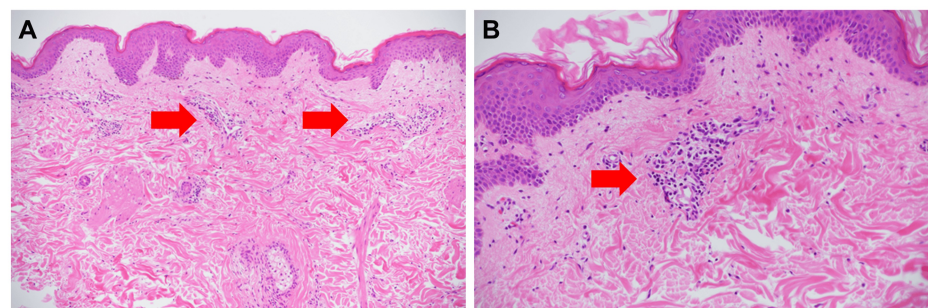


FIGURE 3: Histopathological findings of the erythematous lesion on the trunk

Hematoxylin and eosin staining showed a superficial perivascular inflammatory cell infiltrate in the dermis, predominantly composed of lymphocytes. (A) Low-power view at ×10 magnification and (B) high-power view at ×20 magnification.

Following initiation of prednisolone, the patient showed slight improvement in respiratory status, partial improvement of the pulmonary infiltrates on chest radiography, and resolution of the erythema. The prominent cutaneous findings complicated the diagnostic process. Although nonspecific erythematous eruptions, including worsening facial erythema and erythema resembling a shawl sign, were present, no pathognomonic cutaneous manifestations of dermatomyositis were observed, and dermatomyositis-specific or related antibodies were negative; therefore, a diagnosis of dermatomyositis was not established, and further escalation of immunosuppressive therapy was deferred.

On hospital day 8, the diagnosis was reconsidered, and urinary *Legionella* antigen testing was performed because of the patient's disorientation, hyponatremia, and hepatic dysfunction. The test was positive,

establishing the diagnosis of Legionella disease. The patient denied any recent international travel or use of hot springs or whirlpool baths.

Levofloxacin 500 mg once daily was initiated immediately after the diagnosis, and his respiratory condition rapidly improved. Prednisolone was discontinued after the diagnosis of Legionella disease.

Levofloxacin was administered for a total of 14 days and then discontinued. His clinical symptoms completely resolved, his mobility progressively improved, and he became independent in activities of daily living. On hospital day 19, follow-up laboratory testing showed improvement in the initial laboratory abnormalities, including a serum sodium level of 138 mEq/L, an aspartate aminotransferase level of 24 U/L, an alanine aminotransferase level of 25 U/L, a lactate dehydrogenase level of 139 U/L, a ferritin level of 713.1 ng/mL, and a C-reactive protein level of 0.339 mg/dL. Follow-up chest imaging showed resolution of the pulmonary infiltrates and left pleural effusion. He was subsequently discharged from our hospital and transferred back to the original psychiatric hospital to resume treatment for alcohol dependence.

Discussion

In this case, prominent cutaneous manifestations accompanied by rapidly progressive pulmonary involvement led to consideration of dermatomyositis in the differential diagnosis. However, the findings supporting this diagnosis were limited. The patient did not have pathognomonic cutaneous manifestations, such as Gottron papules or heliotrope rash, and the creatine kinase level was not significantly elevated. Although generalized muscle weakness was observed clinically, precise assessment of muscle strength was limited by his disorientation. Thus, dermatomyositis remained a diagnostic consideration, although the available clinical findings were insufficient to confidently support the diagnosis.

Although Legionella disease is primarily recognized as a cause of severe atypical pneumonia, it may also present with multisystem extrapulmonary manifestations, including gastrointestinal symptoms, hepatic dysfunction, acute kidney injury, and central nervous system involvement. In contrast, cutaneous manifestations are uncommon, with maculopapular eruptions representing the most frequently reported pattern [2]. Nevertheless, the spectrum of skin manifestations is broad and includes erythematous macules, nodules, abscesses, cellulitis, ulcers, bullae, pustules, and panniculitis [4]. Erythema multiforme (EM)-like cutaneous manifestations have also been reported in patients with Legionella infection [5]. Histopathological findings are similarly heterogeneous, including dermal edema with hemorrhage, mild spongiosis with focal parakeratosis, fibrin thrombi, fibrinoid changes of vessel walls, spongiotic microvesicles, and perivascular lymphocytic infiltration [6]. In the present case, the skin biopsy showed superficial perivascular inflammatory cell infiltration predominantly composed of lymphocytes, a finding that has also been reported among the heterogeneous histopathological features of cutaneous manifestations associated with Legionella infection [6]. Although this finding is nonspecific, it is compatible with the spectrum of inflammatory changes reported in Legionella-associated skin lesions.

Reported cases suggest that cutaneous lesions usually appear after the onset of systemic symptoms, either as the illness progresses or during hospitalization after initiation of antibiotic therapy [6]. The underlying mechanism remains unclear, although several hypotheses have been proposed, including direct effects of Legionella-derived toxins, host immune-mediated responses, and drug-induced reactions [6-9]. A dysregulated systemic inflammatory response triggered by Legionella infection or its treatment may therefore contribute to these dermatologic manifestations. In the present case, the precise onset and evolution of the skin lesions could not be confirmed because of the patient's disorientation, and he had received multiple antibiotics before transfer to our hospital. Therefore, a drug-related eruption cannot be completely excluded.

Several host factors have been associated with an increased risk of Legionella disease and poor clinical outcomes, including older age, male sex, cigarette smoking, chronic lung disease, diabetes mellitus, chronic kidney disease, excessive alcohol consumption, and immunosuppression [10,11]. These factors may impair host defense against *Legionella spp.* and increase susceptibility to severe infection. In the present case, the patient had several recognized risk factors for Legionella disease, including male sex, cigarette smoking, and alcohol dependence. These factors, together with severe pneumonia and extrapulmonary manifestations, should prompt clinicians to maintain a high index of suspicion for Legionella disease, even when atypical features such as erythema suggest an alternative diagnosis.

The markedly elevated ferritin level also contributed to the diagnostic complexity. Marked hyperferritinemia can occur in severe infections, including Legionella infection, as well as in autoimmune and hyperinflammatory conditions, including dermatomyositis [12]. Therefore, the ferritin level of 15,600 ng/mL was not sufficiently specific to distinguish between these possibilities.

Although respiratory status and pulmonary infiltrates showed slight improvement and the erythema resolved after initiation of glucocorticoid therapy, antimicrobial therapy was administered concurrently. Glucocorticoids can suppress inflammation nonspecifically regardless of the underlying cause; therefore, a partial clinical response to glucocorticoid therapy should not be interpreted as evidence of an autoimmune

etiology or as a definitive therapeutic response. In this case, the positive urinary *Legionella* antigen test and rapid clinical improvement following initiation of levofloxacin provided stronger evidence supporting the diagnosis of *Legionella* disease.

In the present case, routine blood cultures and sputum cultures were performed; however, neither *Legionella*-specific culture using buffered charcoal yeast extract (BCYE) agar nor polymerase chain reaction (PCR) testing was performed, limiting further microbiological confirmation beyond the positive urinary antigen test. Although the urinary antigen test is useful for the rapid diagnosis of *Legionella* disease, commercially available assays primarily detect *Legionella pneumophila* serogroup 1 and may not detect other serogroups or *Legionella* species [13,14]. Culture of respiratory specimens on selective media, such as BCYE agar, remains the gold standard for diagnosing *Legionella* infection because it can detect a broad range of *Legionella* species and serogroups and provide isolates for epidemiological investigations, whereas molecular testing of respiratory specimens, including PCR and other nucleic acid amplification tests, offers a rapid and sensitive complementary diagnostic approach but cannot reliably distinguish between viable and nonviable bacteria [13,14]. Direct fluorescent antibody testing has limited sensitivity, whereas serological testing is generally of limited value in acute diagnosis because antibody responses may take several weeks to develop [15,16]. Therefore, urinary antigen testing should ideally be complemented by respiratory specimen culture and molecular testing.

Conclusions

Legionella disease may occasionally present with atypical cutaneous manifestations, posing a diagnostic challenge and leading to consideration of autoimmune or inflammatory disorders such as dermatomyositis. Careful assessment of the morphology and distribution of skin lesions is important for differentiating between these conditions. In patients with severe pneumonia and unexplained extrapulmonary findings, appropriate testing for *Legionella* should be considered, particularly when recognized clinical risk factors are present.

Additional Information

Author Contributions

All authors have reviewed the final version to be published and agreed to be accountable for all aspects of the work.

Concept and design: Nanase Honda, Takuto Ishida, Taiju Miyagami, Megumi Shimada

Acquisition, analysis, or interpretation of data: Nanase Honda, Takuto Ishida, Taiju Miyagami, Megumi Shimada

Drafting of the manuscript: Nanase Honda, Takuto Ishida, Taiju Miyagami, Megumi Shimada

Critical review of the manuscript for important intellectual content: Nanase Honda, Takuto Ishida, Taiju Miyagami, Megumi Shimada

Supervision: Nanase Honda, Takuto Ishida, Taiju Miyagami, Megumi Shimada

Disclosures

Human subjects: Informed consent for treatment and open access publication was obtained or waived by all participants in this study. **Conflicts of interest:** In compliance with the ICMJE uniform disclosure form, all authors declare the following: **Payment/services info:** All authors have declared that no financial support was received from any organization for the submitted work. **Financial relationships:** All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. **Other relationships:** All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

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