Case Report

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A Case of Lyme Disease with Parotitis

N. Kawagishi[a]
H. Takahashi[b]
Y. Hashimoto[b]
K. Miyamoto[b]
H. Iizuka[b]

[a] Division of Dermatology,
Kitami Kobayashi Hospital, Kitami,
[b] Department of Dermatology,
Asahikawa Medical College, Asahikawa, Japan

Key Words
Lyme disease
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Abstract
A-59-year-old Japanese woman presented a cellulitis-like erythematous skin rash, low-grade fever, and general fatigue, accompanied by a firm swelling of the right parotid gland. She had a history of tick bite on the right lateral neck 2 weeks before. Serum anti-*Borrelia burgdorferi* antibody was positive by Western blot analysis, and *B. burgdorferi* was isolated from the skin lesion. Serum amylase level was elevated with predominant salivary gland isozyme; the level returned to normal within 3 weeks following penicillin and tetracycline treatment. Parotitis might be included among the rare complications of Lyme disease affecting the head and neck region.

Introduction

Lyme disease is an infectious disease caused by *Borrelia burgdorferi* sensu lato, and transmitted in Japan mainly by *Ixodes persulcatus* ticks. It usually begins with a skin lesion called erythema chronicum migrans (ECM). Weeks to months later, the lesion may be followed by neurologic or cardiac abnormalities, migratory polyarthritis, intermittent attacks of oligoarticular arthritis, or chronic arthritis. It has been reported that Japanese Lyme disease has a relatively milder course, with its cardinal symptom represented by ECM [1]. The most common extracutaneous findings in Japan are arthralgia, fever and headache. ECM typically begins at the site of a tick bite as an erythematous papule that gradually enlarges, assuming an annular configuration. The center of the lesion usually clears but may remain erythematous, indurated, or rarely become necrotic, suggesting cellulitis [2]. We report a case of Lyme disease with a cellulitis-like skin lesion accompanied by ipsilateral parotitis.

Case Report

The patient is a 59-year-old Japanese woman who noticed a tick bite on the right of her neck. Two weeks later, an erythematous rash developed on her right lateral neck and it gradually increased to reach 8 cm in diameter. The skin lesion was associated with necrosis, vesicles, edema, and a burning sensation which looked like cellulitis (fig. 1). During the next 2–3 days, she experienced low-grade fever and general fatigue. This was accompanied by bilateral cervical lymphadenopathy and a firm swelling of the right parotid gland. Laboratory tests showed the following abnormal findings: total peripheral blood leukocyte count, 9,100/μl, with 79.5% neutrophiles, serum amylase level 1,473 units (normal 70–220), with salivary gland-type isozyme 83.6% (normal 40–70). Serum lipase was within normal limits. C-reactive protein was raised at 11.9 mg/dl (normal <0.5). Histopathological examination of the center of the erythema with small vesicles revealed the following features; necrosis of the epidermis, marked edema of the upper dermis, a superficial neutrophilic infiltrate with partial abscess formation and deep perivascular and interstitial infiltrate consisting mainly of lymphocytes, scattered eosinophils and plasma cells (fig. 2). A small site of necrosis of collagen fibers was also found. There were lymphocytic infiltrates around hair follicles and eccrine glands. A Warthin-Starry silver stain was positive for spirochete consistent with the presence of Borrelia. Culture of the patient's skin specimen was negative for bacteria, but the culture in Barbour-Stoenner-Kelly II medium for 5 weeks isolated *B. burgdorferi* sensu lato. Serological tests for *B. burgdorferi* by Western blotting was also positive. She was treated with an intravenous injection of ticarcillin sodium (2 g/day) for 10 days and oral administration of tetracycline (750 mg/day) for 14 days. All symptoms resolved in 10 days following the treatment. The serum amylase level returned to normal after 3 weeks.

Discussion

Lyme disease is a multisystemic disorder caused by the spirochete, *B. burgdorferi* sensu lato. The clinical manifestation can be classified into early localized disease (ECM), early disseminated disease (secondary ECM, arthralgia, meningitis, neuritis, and carditis),

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Yoshio Hashimoto
Department of Dermatology
Asahikawa Medical College
4-5-3-11 Nishikagura 078 (Japan)
Tel. +81 166 68 2523, Fax +81 166 65 7751

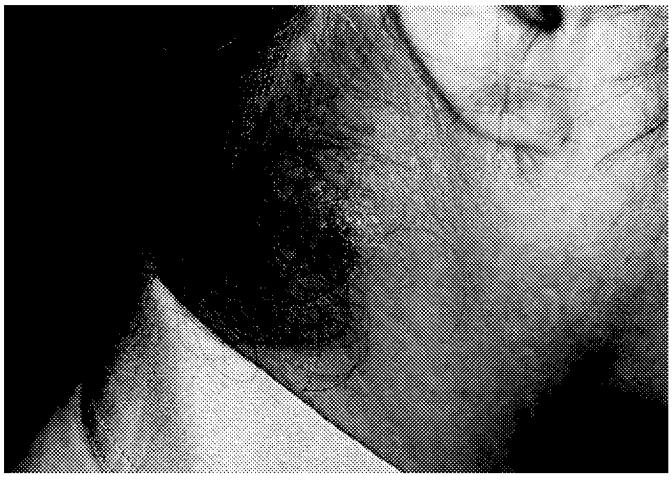

Fig. 1. Erythematous skin rash simulating cellulitis. Center of the lesion shows necrosis, vesicles and edema.

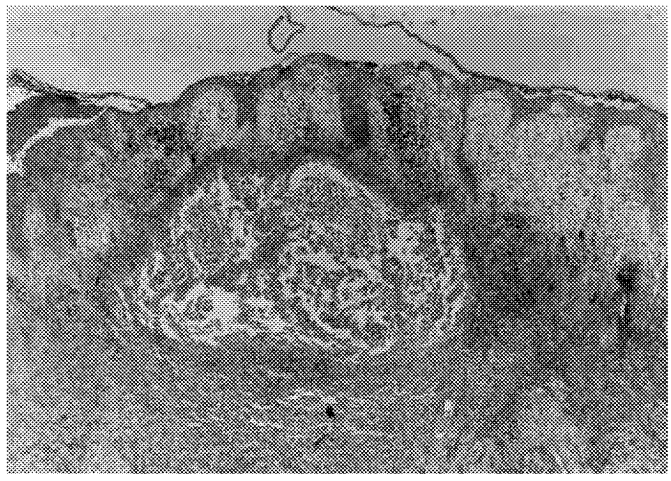

Fig. 2. Low-power view of the lesion. Necrosis of epidermis, marked edema of papillary dermis and a superficial neutrophilic infiltrate with partial abscess formation are noted. HE. ×100.

and late persistent disease (chronic arthritis, acrodermatitis chronica atrophicans, and encephalomyelitis [3]. In Japan, ECM is the main clinical manifestation [1]. During the early phase of the disease, a variety of symptoms, such as fatigue, lethargy, headache, fever, stiff neck, arthralgia, myalgia and lymphadenopathy, may be observed [4]. The otolaryngologic manifestations of Lyme disease include headache, neck pain, odynophagia, cranial nerve palsy, head and neck dysesthesia, otaliga, tinnitus, hearing loss, vertigo, temporomandibular pain, lymphadenopathy, and dysgeusia [5]. However, parotid gland involvement has not been well established. Goldfarb et al. [6] reported a case of Lyme disease with a firm swelling of the parotid gland, but no serum amylase and isoamylase analyses were performed. Our patient had a tick bite on the right lateral neck followed by cellulitis-like erythema migrans (EM) that was accompanied by a firm swelling of the ipsilateral parotid gland. The elevated serum amylase in our patient was of salivary gland origin. A close relation between the tick bite site and the other clinical manifestations has been well documented in Lyme borreliosis [1]. For example, facial nerve palsy is found after tick bite on the ipsilateral auricle [1]. Interestingly facial nerve palsy (neuroborreliosis) has occasionally been reported in Japan [1, 7]. Since the facial nerve runs through the parotid gland, we suggest that the parotid gland could also be a target of Lyme disease. This might occur without evidence of clinical facial nerve palsy.

Our patient showed ECM which simulated cellulitis. Although ECM is the typical cutaneous manifestation of Lyme borreliosis, the diagnosis is not always clear-cut. Paparone et al. [2] described that the classic form of EM with an indurated annular erythematous border is not necessarily pathognomonic. Instead of central clearing, scaling, necrosis or induration sometimes occur. Also the center of the lesion is occasionally erythematous, vesicular or crusted. Feder and Whitaker [8] stressed the pitfalls associated with the diagnosis of erythema migrans that may result in overdiagnosis or underdiagnosis. The cellulitis-like tender erythematous lesion in the present case could be differentiated from bacterial cellulitis by negative bacterial culture and by the clinical course of erythema with a rapidly expanding border. Our case indicates that parotid gland involvement might be among the rare manifestations of Lyme borreliosis. This could be associated with more severe skin lesions on the head and neck which may simulate cellulitis.

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