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Case Report

Chikungunya Presenting as a Bimodal Illness with Atypical Rash

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ABSTRACT

Chikungunya, is now endemic in tropical Asia, Sub-Saharan Africa and the Americas. We describe a case of chikungunya infection presenting as a bimodal illness of fever, rash, arthralgias and arthritis with rash atypically prominent on the palms and soles. This case demonstrates the need for vigilance regarding atypical presentations of chikungunya infection.

INTRODUCTION

Chikungunya Virus (CHIKV) was first identified during an outbreak of febrile illness in 1952 in Southern Province, Tanganyika Territory – present day Tanzania [1]. CHIKV is endemic to the Indian Ocean region, neighboring Asian countries, sub-Saharan Africa [2], with numerous reports of infection in travelers to these regions [3,4]. Autochthonous infection occurring in the Americas was first documented in Saint Martin in 2013 was rapid spread throughout the Caribbean basic and northward to Florida [5]. CHIKV infection is typified by the abrupt onset of fever, headache, fatigue, joint swelling and pain that may be accompanied by a generalized rash involving the trunk and extremities [6]. Less frequent manifestations include encephalitis, involvement of the cardiovascular, renal, and pulmonary systems [6] and hemorrhagic complications [4]. Atypical dermatologic presentations that mimic Stevens-Johnson syndrome or toxic epidermal necrolysis, and palmoplantar erythema have been observed [7]. The rash and fever typically resolve within 7-10 days after disease onset, although joint symptoms may continue for weeks to months.

Here we describe a case of CHIKV infection presenting as a bimodal illness of fever, joint pain and generalized rash involving the palms and soles. This case highlights the need for awareness of atypical presentations of CHIKV infection.

CASE DESCRIPTION

A 27 year-old physician presented in July 2013 with recurrent fever, joint pain, myalgia and a generalized maculopapular rash that was prominent on the palms and soles. She had recently returned from a 5-week trip to Bangalore, India. Symptoms began 16 days prior to presentation, when she developed right knee discomfort associated with fever to 40°C. Fever continued with joint pains extending to the small joints of the hands and feet associated with swelling and acral erythema. On day three, she developed a non-pruritic generalized, maculopapular rash that involved the palms and soles. Diagnostic evaluation in India including malaria smear, leptospirosis

serologies, and dengue serologies were negative. She returned to the United States (US) on day four of illness.

Initial medical evaluation in the US on day 6 of illness revealed a white cell count of 4,200 with a normal differential count, hemoglobin of 11.7, and platelet count of 267,000. Blood chemistries were unremarkable. Serologic testing for mononucleosis, parvovirus, and Lyme disease were negative. Serology against Salmonella typhi O was positive, without a history of a diarrheal or gastrointestinal illness during travel. She was diagnosed with a non-specific viral illness and treated symptomatically. The fevers and rash slowly resolved, with only mild joint pain and swelling and generalized fatigue continuing as of day 10.

On day 14 there was recurrence of fever to 40°C and worsening joint pains, and on day 16 recurrence of a non-pruritic, generalized maculopapular rash that again was prominent on the palms and soles. She was referred for infectious disease consultation. Review of systems was otherwise remarkable only for headaches during peaks of fever. Physical examination demonstrated tender cervical, supraclavicular, and preauricular lymph nodes. There was swelling, tenderness and erythema of the metacarpal and interphalangeal joints of the hands and metatarsal and interphalangeal joints of the feet; as well as anon-blanching generalized maculopapular rash that involved the torso, extremities and included the palms and soles (Figure).

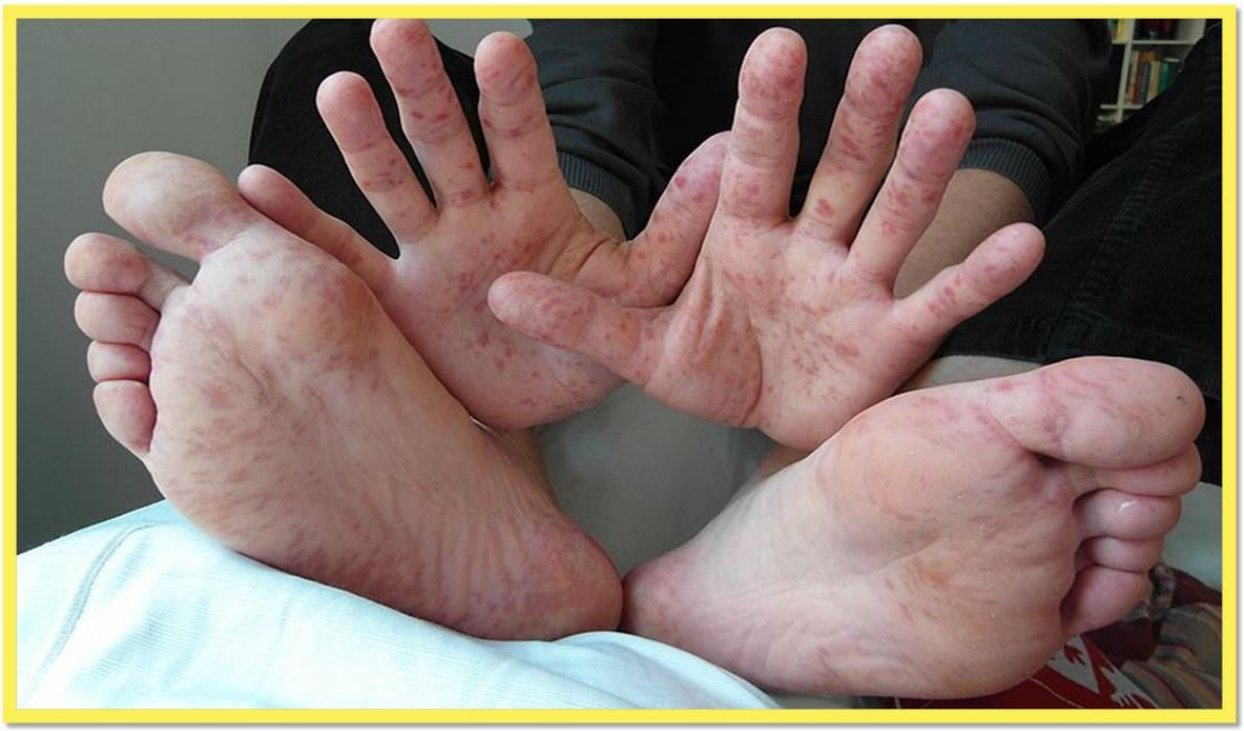

Figure 1: Photograph of the patient on day 17 of illness demonstrating an exanthema involving the palms and soles.

A review of risks for infection noted no ill contacts. There was no travel outside of the United States for the prior 18 months. During her trip to India, she resided with her parents in Bangalore with day trips to non-rural tourist sites. Rats and mice were common in the vicinity of her parent's residence but not in the home itself. She sustained multiple mosquito bites while in India and did not use mosquito repellant. Meals were vegetarian. Of note, there was an outbreak of leptospirosis in India during the time of travel (late May through June), and both dengue and malaria were also being reported in India during the time of travel, although disease activity was not particularly prominent.

Laboratory evaluation was remarkable only for a mild elevation of ALT to 75. Serologic titers for measles, mumps and rubella demonstrated immunity consistent with prior immunization. Repeat testing for parvovirus, leptospirosis, and

dengue were negative. Because of travel to India, CHIKV serologies were performed by a diagnostic reference laboratory on day 16 of her illness that demonstrated an IgM>1:320 and IgG> 10,200, considered indicative of acute CHIKV infection. Symptomatic management was continued. The rash resolved after 10 days (day 26 of her illness) while the arthralgia, joint swelling, and asthenia persisted for approximately three months.

DISCUSSION

This case has two unusual clinical features: a bimodal illness and a generalized rash that was prominent on the palms and soles. A bimodal pattern of disease has to our knowledge not been previously reported for CHIKV infection and was specifically mentioned as not occurring in a case series of 47 travelers returning to France by Simon et al., [4]. Rash involving the palms and soles has been infrequently reported. The report by Simon et al. noted the occurrence of rash and tenosynovitis of the hands in approximately half of patients but without exanthem involving the palms or soles [4]. Hochedez et al., reported on 22 French travelers returning from the Indian Ocean region of whom 3 had rash involving the palms and soles [3]. Rahim and Udin reported on 7 patients in Bangladesh with CHIKV infection, of whom 2 reported rash of whom one case involved the palms and soles [8]. Riyaz et al., reported on 163 patients from Calicut, India of whom 2 infants developed palmar pigmentation and 10 adults who developed palmoplantar desquamation without mention of either adults or infants developing an exanthem involving the palms or soles [9]. Two other Indian case series from 2006 of 115 patients and 75 cases from June-August 2008 with clinically diagnosed CHIKV describe swelling of the hands and feet and palmo-plantar desquamation in a small number of patients but without exanthem of the palms or soles [10,11]. Interestingly, rash may be delayed up to a month after the acute illness has resolved [12].

Diagnostic uncertainty was apparent in this case both at the time of onset in India and again upon initial evaluation following return to the US. At the time of travel in 2013, CHIKV was not prevalent, but was endemic in India at that time. Additionally, the known outbreaks of leptospirosis, dengue, and malaria in India may have resulted in diagnostic bias. Other alphaviruses such as Venezuelan equine encephalitis, eastern equine encephalitis, western equine encephalitis, or Mayaro virus were not prevalent in the patient's region of travel or residence nor were there relevant epidemiology exposures. The finding of a high value of IgM antibodies against CHIKV was considered diagnostic.

Due to the global prevalence of CHIKV, vigilance for atypical presentations is important for timely diagnosis. Attention to bimodal pattern of disease, unusual dermatologic manifestations.

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