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Case Report

Gianotti-Crosti syndrome: exanthema following a vaccination

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ABSTRACT

Gianotti-Crosti syndrome (GCS) is a benign skin disorder characterized by self-limiting acute exanthema. Commonly preceding with the infection of viruses or bacteria and vaccinations. Usually affects children aged 1-6 years and is associated with atopic disease. The exact of GCS incidence is unknown, although several countries in the world have been reported GCS cases. Vaccine-induced GCS cases are infrequently reported. The diagnosis can be made clinically based on clinical features and vaccination history. Treatment is generally symptomatic. Education about GCS is important to prevent over-treatment and avoidance of vaccination in the future. We reported a 9-month-old baby with symmetrical bilateral monomorphic multiple dome-shaped papules lesions on the extremities two days following vaccination.

Keywords: Gianotti-crosti syndrome, Exanthema, Vaccination

INTRODUCTION

Gianotti-Crosti syndrome (GCS) or papular acrodermatitis of childhood is an acute self-limiting exanthema that usually occurs in childhood.[1-3] Generally affects children aged 1-6 years. Adult cases are rarely reported and slightly more in the woman. GCS usually begins with infections (viral or bacterial) and vaccinations. The delayed immune response (type 4 hypersensitivity reaction) is presumed to have an important role in this disease.[1,3,4] Lesions are usually found on the extremities of the extensor area, buttocks, and face.[1,2] GCS was first reported in 1953 by Fernando Gianotti, an Italian pediatric dermatologist together with Professor Agostino Crosti in Milan as monomorphic erythematous lesions on face and extremities in infants and children.[1,4] In general, the incidence of GCS is unknown although many GCS cases are believed to be underdiagnosed as viral rash or nonspecific viral exanthema.[5,6] Nonetheless, many countries such as Great Britain, France, Germany, Spain, Russia, Turkey, Hong Kong, China, Japan, India, and Singapore have been reported GCS cases.[7-8] We reported a GCS case in a 9-month-old baby following a vaccination.

RESULTS

A 9-month-old baby girl complained of having multiple papules on both forearms, legs, and buttocks four days ago. Initially, the skin lesions appear on the buttocks then spread to the forearms and legs. One week ago, the patient had just received the measles and rubella (MR) vaccination at the public health center and the next day the patient suddenly had a fever of 37.7 ℃. The mother thought her child had an adverse event following immunization (AEFI) reaction and gave paracetamol to her baby. The fever disappeared with one dose administration of paracetamol but the next morning there were papules on the buttocks who within two days spreading to her forearms and legs. Since the skin lesion appeared, the mother said that her child was fussy and wakeful. The patient is first child, full-term, born with cesarean section. No natal or antenatal complication. The mother has a history of asthma and the father has a history of cat dander

allergy. A history of similar complaints in the family, having respiratory or digestive tract disorder before skin lesion appeared, and hepatitis disease in parents was denied.

On physical examination, the baby appears healthy. Fever, lymphadenopathy, hepatomegaly, and splenomegaly were not found. Dermatologic examination revealed bilateral symmetrical distribution, multiple, flesh to erythematous dome-shaped papules, mostly discrete, 1-5 mm on wrists, knees, and dorsum pedis (Figures 1 and 2). Complete blood count and liver function were normal. Hepatitis B virus serology was negative.

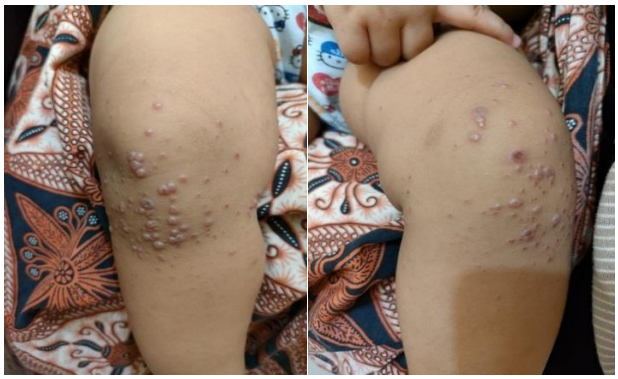

Figure 1: Bilateral symmetric, multiple dome-shaped papules monomorphic with flesh to erythematous color on right and left of knees.

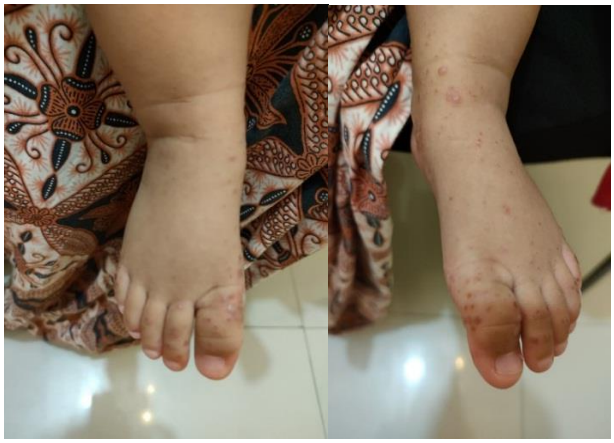

Figure 2: Bilateral symmetric, multiple dome-shaped papules monomorphic with flesh to erythematous color on right and left of dorsum pedis.

The GCS diagnosis was made clinically. The patient has received a mid-strength topical corticosteroid (mometasone furoate 0.1% cream) applied twice daily for one- week, emollient moisturizer, and systemic antihistamine. Skin lesions completely healed without scarring after 2 weeks of treatment. Initially, the mother worried for doing vaccinating in the future, then we did an explanation and education the disease is benign and usually spontaneously improve. So, there is no need to hesitate for revaccinating her child later.

DISCUSSION

GCS is an acute exanthema with benign and self-limiting dermatosis. GCS can affect children within 3 months to 15 years old, with a peak aged 1 until 6 years. In adults GCS commonly affects women, there is probably hormonal involvement. GCS was usually associated with atopic disease.[1-3] At first, the hepatitis B virus was considered to be the main cause due to high GCS cases found in anicteric hepatitis patients. But now in worldwide, Epstein-Barr Virus (EBV) infection is the most common cause in GCS. Many other viral infections can lead to GCS such as cytomegalovirus, hepatitis A and C viruses, Mumps virus, Rubella virus, HHV-6, herpes simplex virus, human immune-deficiency virus (HIV), poxvirus, poliovirus, respiratory syncytial virus, influenza virus, adenovirus, rotavirus, and enterovirus. Bacterial infections such as *M. pneumoniae*, *Borrelia burgdorferi*, *Bartonella henselae*, and group A *Streptococcus*. In addition, many GCS cases preceded by vaccination have also been reported.[1,8-10] Vaccines associated with GCS are as following *Haemophilus influenza* type b (Hib); oral polio; diphtheria, pertussis, and tetanus (DPT); measles, mumps, and rubella (MMR); bacillus Calmette-Guerin (BCG); hepatitis B and A, and Japanese encephalitis.[1,5,9,11] It was reported in the literature that 21 out of 300 GCS cases were associated with vaccination.[9]

There are two main hypotheses of GCS pathogenesis. First, the immune response is mediated by immunoglobulin E (IgE), it is supported by elevated serum IgE in patients with GCS where usually also have a history of atopic disease. Second, the delayed hypersensitivity reaction (type 4 hypersensitivity reaction) in the dermis layer as a response to infections (virus or bacteria) and vaccines.[1,4,5] Another study by Caltabiano et al reported that discover increasing human beta-defensin-4 (hBD-4) activity in the epidermis, indicating viral antigenemia probably has a role in pathogenesis of GCS.[12]

On general examination, the child usually appears healthy. GCS is characterized by acute onset of monomorphic dome-shaped or flat-topped papulovesicular lesions, in varying size from 1 mm to 5 mm, flesh color to reddish-brown. Symmetrical distribution on the extensor surface of the extremities, buttocks, and face. The lesions may coalesce and form a larger plaque. Hemorrhagic or scaly lesions are uncommon.[1-3] Lesions are infrequently affecting the trunk, palmar or plantar, and mucosa, but several studies have been reported atypical manifestations of this area. The initial location is usually on the buttocks and spreads to other locations. Skin manifestations may be preceded by malaise, low-grade fever, and lymphadenopathy (general, cervical, axillary, and inguinal). Hepatomegaly is associated with hepatitis B infection while splenomegaly is associated with EBV infection.[1,2,4]

The GCS diagnosis can be made clinically. Laboratory tests or skin biopsy are generally not required. The biopsy

may be considered if the skin lesions persist for more than 6 months or GCS cases with atypical presentation and exclude the differential diagnosis in high-risk or immunocompromised patients.[1,2,10] Histopathological features are nonspecific showed parakeratosis of the epidermis, focal spongiosis, mild acanthosis, and psoriasiform epidermal hyperplasia. Perivascular lymphohistiocytic infiltrate with scattered eosinophils and extravasation of erythrocytes in the dermis.[1,2] If on examination were found hepatosplenomegaly or suspected a hepatitis infection then viral serologic examination, complete blood count, and liver function test should be performed to find the underlying causes.[1] Differential diagnoses of GCS is erythema infection or another viral exanthema, id reaction, urticaria papular, hand, foot, and mouth disease, Langerhans cell histiocytosis, scabies, and mononucleosis infectious.[1,2]

GCS does not require specific therapy due to benign and self-limiting courses. Treatment is usually symptomatic. Several studies have been reported that using emollient moisturizers and topical or oral antihistamines can help relieve symptomatic complaints. Mild to moderate potency of topical corticosteroids may be administered if the itching does not improve with emollients and antihistamines. If any underlying cause of GCS such as hepatitis B also should be treated. The lesions usually fade within 10 days to 3 months without scarring, although some cases cause hyperpigmentation and hypopigmentation post-inflammatory in darker skin types. Some lesions can persist for 6 to 12 months. Our patient responded well to moderate potency of topical corticosteroid, emollients, and antihistamines within 2 weeks on treatment.[1,2,5,9]

The prognosis of GCS is generally good, recurrence is unusual. There were 3 relapse cases have been reported in the literature. Assaf et al reported a recurrence case following 2 different types of vaccination: vaccinated with hepatitis B and 3 months later having recurrence after receiving MMR vaccine.[5] On the other hand, two cases were reports that administration of a single episode MMR vaccine does not associate with GCS cases recurrence.[5,9] Education to the parents about GCS is important to prevent unnecessary tests and treatment also vaccination avoidance later.[5,6,11] Parents of our patient also have the same condition where they worried to give vaccination again to their child. This is a new challenge for reassuring parents that vaccination is not a contraindication in the future due to GCS being a benign self-limiting disease.

CONCLUSION

Clinical features such as the onset, duration, distribution, color lesion, and vaccination history can aid to establish the GCS diagnosis and rule out the differential diagnosis. GCS recurrence by the vaccine is less common but has been reported in several studies. GCS is not a vaccination contraindication in the future.

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