

Acute hemorrhagic edema of infancy after bacillus Calmette-Guerin vaccination

Rajaa Bousmara ^{1,*}, Fatima Ezzahra El Fatoiki ¹, Fouzia Hali ¹, Farida Marnissi ² and Soumiya Chiheb ¹

¹ Department of Dermatology-Venereology, UHC Ibn Rochd Hassan II University, Casablanca, Morocco.

² Department of Anatomical Pathology, UHC Ibn Rochd, Hassan II University, Casablanca, Morocco.

World Journal of Advanced Research and Reviews, 2025, 26(03), 1661-1663

Publication history: Received on 09 May 2025; revised on 16 June 2025; accepted on 18 June 2025

Article DOI: <https://doi.org/10.30574/wjarr.2025.26.3.2373>

Abstract

Acute hemorrhagic edema of infancy (AHEI) is a rare and benign form of leukocytoclastic vasculitis that usually affects children younger than 2 years, and is often misdiagnosed by pediatricians and dermatologists. Numerous precipitating factors have been reported, including drugs, infections, and vaccination. We present herein a unique case of AHEI following bacille Calmette–Guérin (BCG) vaccination. To our knowledge, this represents the second reported case after BCG vaccination.

Keywords: Hemorrhagic edema; Vasculitis; BCG vaccine; Infancy

1. Introduction

Acute hemorrhagic edema of infancy (AHEI) is a rare and benign form of leukocytoclastic vasculitis [1]. Numerous precipitating factors have been reported, including drugs, infections, and vaccination [1]. We report the case of a 3-week-old boy with AHEI following the bacillus Calmette–Guérin (BCG) vaccination. To our knowledge, this is the second reported case of AHEI following the BCG vaccine.

2. Case Report

A 3-week-old male infant presented to our dermatological department with a 15-day history of progressive erythematous and indurated plaques with edema on the face and extremities. There was no previous history of infection or use of medications. However, the patient had been vaccinated with the BCG vaccine 3 days before presentation. Physical examination revealed multiple purpuric, annular, and targetoid plaques measuring 1 to 4 cm in diameter, predominantly on the face (Figure 1), and with marked edema on the left eyelid accompanied by a large cockade and purpuric plaque (Figure 2). Similar but smaller lesions were observed on the extremities, while the trunk and back remained unaffected. In addition to this clinical presentation, the patient showed multiple inflammatory papules and pustules without comedones on the face, suggesting benign cephalic pustulosis. There was no involvement of the mucous membranes. The rest of the physical examination showed no systemic symptoms. Skin biopsy revealed leukocytoclastic vasculitis. Direct immunofluorescence (DIF) was negative. Chest X-ray and abdominal ultrasonography were normal. Laboratory blood tests revealed neutrophils measuring 18000/mm³. Renal, liver function and coagulation tests were normal. A diagnosis of AHEI was made. The patient's skin lesions spontaneously improved without treatment over three weeks, and no relapse was observed.

* Corresponding author: Rajaa Bousmara



Figure 1 Maculopapular and purpuric annular lesions on the face



Figure 2 Purpuric targetoid edematous plaques on the left eye

3. Discussion

Acute hemorrhagic edema of infancy (AHEI) is a rare and benign form of leukocytoclastic vasculitis. It is slightly more common in males and usually occurs in children under two years of age [1]. AHEI is characterized by the classic triad of fever, edema, and round, palpable, purpuric plaques predominantly affecting the face, earlobes, and extremities [2]. The skin lesions are typically palpable, asymptomatic, and symmetrically distributed [2]. Histopathological examination reveals leukocytoclastic vasculitis [2]. Prompt recognition of AHEI is crucial for differentiating it from other conditions requiring specific treatment, particularly IgA vasculitis. In our patient, there was no predominant involvement of the extremities or any visceral involvement, and negative staining for IgA on DIF, along with the rapid resolution of the lesions, allowed us to rule out this diagnosis [4]. Considering the annular lesions, neonatal lupus was also evaluated;

however, laboratory findings revealed no leukopenia and both antinuclear and antineutrophil cytoplasmic antibodies were undetectable. The child's stable clinical condition and negative serological tests ruled out recent infections [5].

The pathogenic mechanism is unclear, but AHEI is considered an immune hypersensitivity complex and often follows infections, drug therapy, or vaccination [1]. In this case, the BCG vaccine was probably responsible for the disease. Only a few cases of AHEI after vaccination have been reported, including MMR vaccine, diphtheria–pertussis–tetanus– polio, H. influenza type b, H1N1, d, conjugated pneumococcal vaccines, and hepatitis B [3]. Only one case following vaccination with BCG has been reported [5].

4. Conclusion

Although vaccinations are rarely implicated in the occurrence of this disease, this diagnostic hypothesis should be raised following BCG vaccination. It is important to reassure parents about this post-vaccination effect because it is benign and resolves spontaneously.

Compliance with ethical standards

Disclosure of Conflicts of interest

There are no conflicts of interest.

Statement of informed consent

The patient in this manuscript have given written informed consent to publication of the case details.

Funding

This research received no external funding.

References

- [1] Bronz G, Consolascio D, Bianchetti MG, Rinoldi PO, Betti C, Lava SAG, Milani GP. Köbner and Pastia Signs in Acute Hemorrhagic Edema of Young Children: Systematic Literature Review. *Children (Basel)*. 2022 Feb 15;9(2):265. doi: 10.3390/children9020265.
- [2] Bronz G, Rinoldi PO, Lavagno C, Bianchetti MG, Lava SAG, Vanoni F, Milani GP, Terrani I, Ferrarini A. Skin Eruptions in Acute Hemorrhagic Edema of Young Children: Systematic Review of the Literature. *Dermatology*. 2022;238(3):397-403. doi: 10.1159/000519009. Epub 2021 Sep 22.
- [3] AlSufyani MA. Acute hemorrhagic edema of infancy: unusual scarring and review of the English language literature (2009). *Int J Dermatol*, 48, 617–622. DOI: 10.1111/j.1365-4632.2009.03917.x
- [4] Hikmet Tekin Nacaroglu, Solmaz Saygaz, Özlem Saraç Sandal, Canan Şule Ünsal Karkiner, Hülya Tosun Yıldırım, Demet Can. Acute Hemorrhagic Edema of Infancy after Vaccination: A Case Report (2014). *J Pediatr Inf* 2014; 8: 40-3
- [5] Francesco Miconi ,Lorenzo Cassiani ,Emanuela Savares, Federica Celi, Manuela Papini, Nicola Principi, and Susanna Esposito. Targetoid Skin Lesions in a Child: Acute Hemorrhagic Oedema of Infancy and Its Differential Diagnosis (2019). *Int. J. Environ. Res. Public Health*, 16(5),823. doi: 10.3390/ijerph16050823