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Acute Hemorrhagic Edema with extensive trunk involvement and Rhinovirus infection association

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Abstract

Acute Hemorrhagic Edema (AHE) is a rare form of leukocytoclastic vasculitis with an unknown etiology. There are various possible triggers associated with the development of AHE such as Upper Respiratory Tract related infections which have been reported in the literature. A typical case of AHE is a child less than 2 years of age having the symptoms of fever, Acral Targetoid Purpura, and Edema. The course of this illness is benign with a complete resolution within few weeks. This case report involves a patient having the classical symptoms of fever, Purpura and Edema, but with an unusual extensive truncal involvement. Moreover, Rhinovirus infection testing was positive in the child's case. The presence of the virus gives a suspicion of being the possible trigger for the boy's AHE. In the previous hospital, the treatment started with steroid for a course of one week without a significant improvement which brought the patients to the Emergency department. A decision of continuing a supportive treatment led to a complete resolution. After 3 weeks the AHE symptoms were gone without any complications.

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Keywords: Acute Hemorrhagic Edema; Leukocytoclastic vasculitis; Purpura; Rhinovirus; Case report

1. Introduction

Acute Hemorrhagic Edema (AHE) is a rare benign form of leukocytoclastic vasculitis with unknown etiology (Oliveira et al., 2015). This disease has a higher incidence during the cold months (Sorensen et al., 2014). The main cause of AHE is still questionable. Nonetheless, various associations have been reported such as infections that may play part of its pathogenesis (Fiore et al., 2008). Other

associations have been reported like after using drugs or vaccinations (Freitas and Bygum, 2013; Findal and Kura, 2013). AHE has a male predominance and usually affects children less than 2 years of age. Usually, the affected children have typical triad of fever, Edema of the face and extremities, and purpura of Targetoid or annular pattern, classically called cockade (rosette or iris-like) pattern (Fiore et al., 2008). AHE diagnosis is clinically based with the skin biopsy to confirm the typical leukocytoclastic vasculitis changes (Fiore et al., 2008). Treatment is only supportive, although there are case reports of use of systemic steroid treatment (Sorensen et al., 2014; Fiore et al., 2008; Freitas and Bygum, 2013; Risikesan et al., 2014).

2. Case report

A 21 month old boy presented to the emergency department with a history of low grade fever, swelling of the face

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and arms, and a rash lasting for 2 weeks. The child had coryza and cough that preceded these symptoms by at least 5 days. Upon the appearance of rash, his parents sought for medical advice. The child had treatment for a week with a combination of antibiotic, antihistamine and an oral steroid. The treatment has resulted in a mild improvement, but the patient continued in developing more lesions and his mother decided to visit the emergency department.

In the Emergency room (ER), the child's mother stated that the rash had started as an itchy reddish lesion then it has become darker in time. The fever was documented by the mother which is 38–39°C responding well to paracetamol. Moreover, the child had some swelling in the face and the arms intermittently with no shortness of breath. Three days before visiting the ER he developed diarrhea mucoid with no blood, no abdominal pain or vomiting and no change in the urine. The child has no history of any allergies. His nutritional developmental, and Family history were unremarkable. He had all vaccines up to his age. He was born full-term by cesarian section with no complications.

During the observation of the baby, he looked well, vitally stable with nonpitting edema of arms, cheeks, left lower eyelid and to lesser extent the upper lip. Multiple palpable Targetoid purpuric plaques over the face, neck, shoulders and trunk were apparent with some forming a polycyclic pattern especially on the latter (Fig. 1). Scalp, mucous membranes, palms, soles and nails were unremarkable. There is no lymphadenopathy or organomegaly detected. His mother gave us some pictures taken for initial lesions (Figs. 2–4).

In the ER, the baby was given a dose of Intravenous hydrocortisone and diaphenilhydramine. Patient was

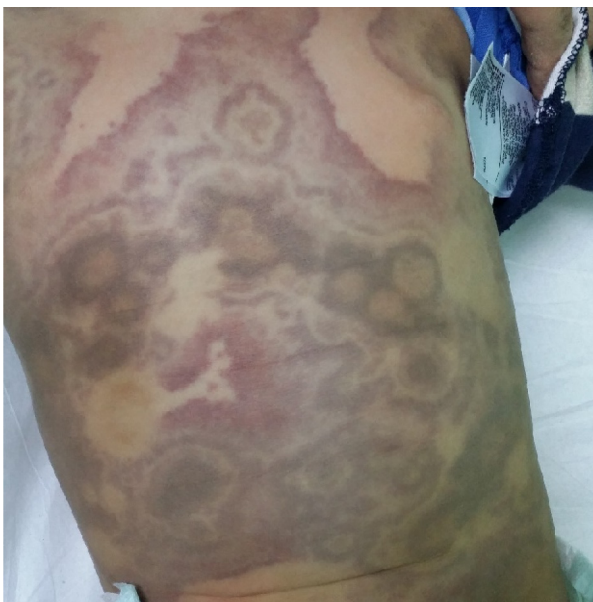


Figure 1. Multiple erythematous to violaceous targetoid purpuric patches that has different shades and coalescing to form a geographic configuration.

admitted with clinical diagnosis of acute hemorrhagic Edema. In the ward, the decision of stopping the steroid was made and only supportive care was continued including antihistamine for the pruritus. As well, blood tests were done and the results show a complete blood count, Erythrocyte sedimentation rate, coagulation profile, kidney function test, liver function test and Anti Nuclear Antibodies, within the normal range. However, there was a slight elevation in Lactate dehydrogenase (LDH) of 357 U/L (normal range 135–225 U/L), elevated C-Reactive protein of 18.1 mg/L (normal range 1–3 mg/L), and IgE level of 272 Ku/L (normal range 1–100 Ku/L). In addition, the infection screening showed positive testing for the Rhinovirus Polymerase Chain Reaction using nasal swab (see shown in Table 1).

For the treatment, an overall supportive care was provided. Within days purpuric lesions started vanishing and normalization of the acute phase reactant (Fig. 5). After 5 days of admission almost all lesions disappeared (Fig. 6).

With clearance of the purpura and the improvement of the patient pruritus, the child was discharged from the hospital with antihistamine as needed and an appointment after 4 weeks. The patient came back at the appointment date having no recurrence.

3. Discussion

The boy was good looking, less than 2 years of age, who had symmetric purpuric targetoid lesions that resolved completely within around 3 weeks and with no internal involvement; it is clear that the diagnosis is consistent with Acute Hemorrhagic Edema. Henoch Schonlein Purpura (HSP) is an important differential diagnosis which is more likely to occur in an older age, and it has a risk of internal involvement. As well, it has a longer duration to resolve, and the relapse has been described (Sorensen et al., 2014). A systematic review on 294 children in a study published in 2008 by Fiore et al suggested criteria for diagnosis: targetoid purpura, nonpitting edema and platelet count of at least $150 \times 10^9/L$. The biopsy is an important tool for confirming diagnosis. Fiore et al describe the typical patient



Figure 2. Erythematous targetoid plaques with one showing a bluish purpuric center.



Figure 3. Edema of the eyelids and erythematous plaques, over lateral face, ears, and neck. The latter show targetoid configuration.



Figure 4. Well-developed targetoid purpuric plaques that is at an earlier stage compared to Fig. 1.

as a boy with age from 6 months to 2 years and history of respiratory infection (Fiore et al., 2008). The unusual symptoms in our patient were the extensive trunk involvement, pruritus and the presence of urticarial wheels, all of which were an uncommon feature as described by Fiore et al. (2008). AHE may show nonspecific changes like thrombocytosis, leukocytosis, elevation of acute phase reactants, and low complement levels (Alhammadi et al., 2013). This type of leukocytoclastic vasculitis is transient with different triggers of which are the virus infections (Fiore et al., 2008). Rhinovirus infection was confirmed with the positive PCR in our patient which may have a causality relationship with Acute Hemorrhagic Edema. Rhinovirus association was reported in the literature with

Table 1
Infections screening done for the patient.

Investigations	Results
Adenovirus PCR	Negative
ASO titer	Negative
Coronavirus PCR	Negative
CMV	Positive(IgM, IgG)
EBV	IgG positive
Mycoplasma	Negative(IgM, IgG)
Rhinovirus PCR	Positive
Rotavirus direct test	Negative
RSV PCR	Negative



Figure 5. Abdomen and thighs showing vanishing of the purpuric eruptions leaving light brown annular patches some having concentric circles as on the thighs.



Figure 6. All lesions cleared.

evidence of coexisting echovirus and coxsackie virus infections in a case report (Oliveira et al., 2015).

For the treatment, supportive care has been the main approach unlike the course of steroids which did not show much improvement in the child's previous treatment. Although systemic steroid therapy has been reported to be beneficial in some case reports, with the benign course

of the illness supportive treatment is considered reasonable treatment (Fiore et al., 2008; Freitas and Bygum, 2013; Risikesan et al., 2014).

4. Conclusion

AHE is a benign form of leukocytoclastic vasculitis that normally needs supportive therapy and clears without any complications. Infections, mainly upper respiratory tract infections, could possibly be involved in development of such disease. In the child's case, Rhinovirus is perhaps the trigger for the AHE condition.

Disclosure

No conflict of interest.

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