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CASE REPORT

# A Case of Acute Infantile Hemorrhagic Edema Confused with Child Abuse

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**ABSTRACT**

Acute infantile hemorrhagic edema (AIHE) is a type of leukocytoclastic vasculitis with a generally good prognosis and is observed in infants aged 4-24 months. It may be difficult to diagnose in some cases because its clinical findings are similar to those of many other diseases. Trauma and child abuse are among the differential diagnoses and should be first excluded. Although the exact cause of this disease is not known, it has been associated with infections, vaccines, and medications. Staphylococci, Streptococcus, and *Adenovirus* are the leading infective agents, while many other agents, such as *Escherichia coli* and Mycobacteria, have been suggested to be the cause of AIHE. Typically, purpuric skin lesions, edema, and fever are the presenting findings. Herein, we report the occurrence of the disease in a 6-month-old girl within a short period after vaccination.

**Keywords:** Acute hemorrhagic infantile edema, Finkelstein's disease, purpura

## INTRODUCTION

Acute infantile hemorrhagic edoema (AIHE) is a rare but generally benign leukocytoclastic vasculitis. This disease is mostly observed in infants (between 4 and 24 months). Clinically, purpuric lesions on the skin, edoema, and fever are common findings. These lesions mostly occur on the cheeks, extremities, and auricles. Lesions usually heal spontaneously, leaving pigmentation. Although the etiology of it is not known, many cases have been reported after infections, vaccines, and some drugs being used (1-5).

Here, we present a case in which disease symptoms appeared within 6 hours of routinely administering combined vaccines [diphtheria-acellular pertussis-tetanus-inactivated polio-haemophilus influenza type b (DaBT-IPA-Hib)], oral polio (OPA), and hepatitis B (Hep-B)] to a 6-month-old girl.

## CASE REPORT

A 6-month-old female patient was admitted to our emergency department with complaints of sudden edoema, redness, and bruising. Within a few hours, new ecchymotic lesions appeared, and they did not fade upon pressure application. First, it was swelling and then bruising; it appeared on the left arm, both sides of the left hand, the left leg, and both sides of the left and right feet, and was painless (**Picture 1a-c**). The family members stated that the patient was vaccinated with DaBT-IPA-Hib, Hep-B, and OPA early in the morning of the same day.

When the patient's medical history was questioned, it was learned that she was born at term and weighed 3,300 g vaginally. The patient was breastfed for the first 2 weeks after birth and then fed supplementary food. No history of medication use or infection before the lesions. No allergic reactions to foods, medications, or vaccines were observed. When the family history was asked, he had no history of similar or other diseases in either of his siblings.

There were no findings other than ecchymotic lesions on physical examination. At first admission, her fever was 37°C, and her pulse rate was 130 beats/min. Initial examinations in our emergency department revealed a white blood cell count of 21,770/mm3 (53.1% neutrophils, 41.8% lymphocytes, 4.2% monocytes, 0.8% eosinophils, and 0.1% basophils), hemoglobin of 8.6 g/dL, and a platelet count of 513,000/mm3. ALT: 12 U/L, AST: 33 U/L, CRP: 22.40 mg/L, INR: 1.13, Prothrombin time: 12.6 seconds. Venous blood gas and complete urinalysis were within normal limits.

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A superficial tissue ultrasound examination revealed mild edematous changes in the subcutaneous tissue in ecchymotic regions with linear fluid echogenicity. Transfontanel ultrasound and fundus examinations performed to exclude possible trauma to the child (child abuse) were normal. The patient, who was observed in our emergency department for a while and underwent relevant tests, was followed up and treated in our allergy and immunology department. The C3 level was: 1.32 g/L, C4 level: 0.30 g/L, ANA level: 1/100 (nuclear-granular) positive, ANCA level: negative, anti-ds DNA level: negative, Rheumatoid factor: 173 IU/L, D-dimer: 5,700 ugFEU/L, ASO: <57.50 IU/mL. We found that rhinovirus/enterovirus was positive in the respiratory system infection panel that we tested. The result of a 3-mm skin biopsy of the ecchymotic lesion on the patient's left arm was consistent with leukocytoclastic vasculitis. The patient we were following in our department was treated with 1 mg/kg/day methylprednisolone and 1 mg/kg/day pheniramine maleate for 5 days. The examination the next day revealed no fever or tenderness, and the lesions had begun to fade. The patient was discharged on the 6th day of hospitalization, and the ecchymotic lesions almost completely disappearing. (The patient gave her consent for this presentation.)

**DISCUSSION**

AIHE, also known as Finkelstein-Seidlmayer vasculitis or Seidlmayer purpura, was first described in 1913 by Snow in an article titled 'Purpura, urticaria, and angioneurotic edoema on the hands and feet'. According to the reported case series, most of the patients were men aged 2 years (1-4).

Although the exact cause of AHIE is not known, it is believed to be caused by vaccines, medications, and infections (5). The fact that our patient was positive for rhinovirus/enterovirus and the lesions appeared shortly after DaBT-IPA-Hib, Hep-B, and OPA vaccinations supports this view, but it leads to confusion about the real etiological trigger. Krause and his team proposed 4 criteria to help diagnose this disease (6). These criteria are as follows:

1) Purpuric rashes in children below 2 years old

2) Purpuric rashes accompanied by edoema on the face and extremities

3) No systemic or visceral lesions or skin lesions lasting several days to weeks

4) Lesions on the skin taking the shape of a target board within 24 to 48 hours and then regressing spontaneously within days or weeks.

Our patient findings typically met the criteria specified here. However, the ecchymotic lesions on the hands, feet, and forearms were more visible than those in other reported case examples (**Picture 1**).

Although edoema and lesions are frequently seen on the arms, legs, and ears in AIHE, trunk muscle involvement is very rare. Fever may usually accompany the lesions. In our case, edoema was evident on the extremities and ears, but fever was at normal levels at both presentation and during the follow-up period (7-11). In the treatment of our patient, antihistamine and methylprednisolone treatments mentioned in the literature were applied; the lesions regressed within a few days, and the patient's lesions completely resolved within 5 days (**Picture 2**).

The diagnosis is usually made by anamnesis and physical examination. There are no specific laboratory tests. Although sedimentation rate (ESR) and CRP levels are generally within the normal range in patients (12), CRP levels were increased in our patient. Mild lymphocytosis and eosinophilia-dominated leukocytosis can be seen in the hemogram; a slight increase in platelet levels can be seen (4,13,14). However, coagulation tests are generally normal (15). Urinalysis is often within the normal range, but microscopic hematuria and proteinuria have also been detected in some patients (12). Antinuclear antibody (ANA) and rheumatoid factor are generally negative, as in our patient (16). Patients with increased C3 and slightly decreased C4 levels have also been reported (2,17). The laboratory images mentioned were consistent with our patient's results.

When the biopsy material from the purpuric region is examined with a light microscope in these patients, karyorrhexis and purpuric-type vasculitic lesions that mostly involve the vessels of the upper and lower dermis can be seen. Fibrinoid necrosis can also be seen around and inside affected vessels, along with erythrocytes and leukocytes (2). We found that the biopsy results for our patient were consistent with the reported data.

The most frequently confused disease when diagnosing AIHE is Henoch-Schönlein purpura (HSP). Although ecchymosis and

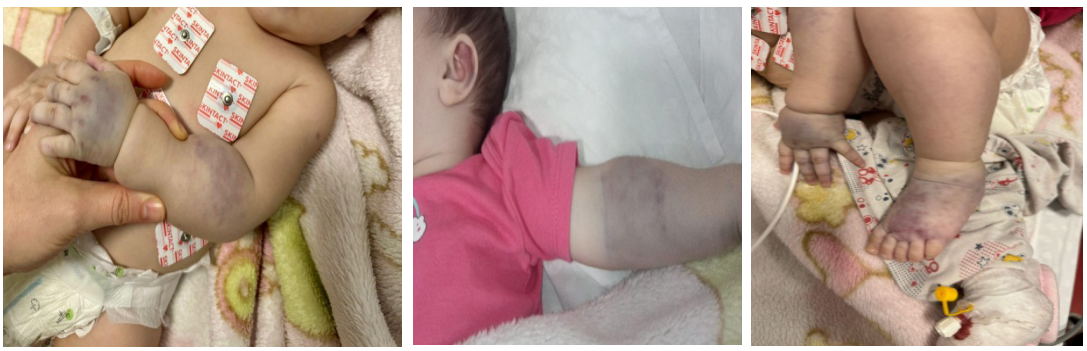

**Figure 1a, b, c: Lesions indicative of trauma at initial presentation and at specific sites.**

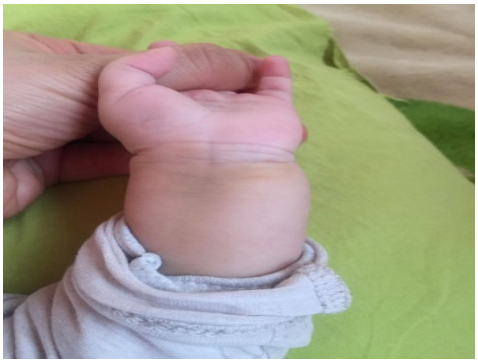
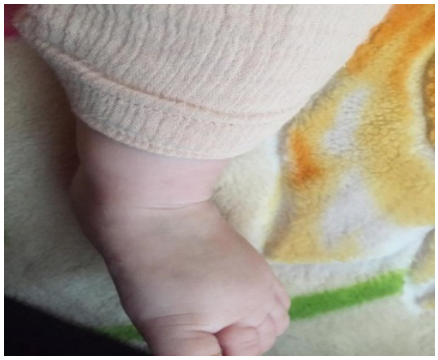

**Figure 2a, b:** Patient's lesions after discharge.

edoema are often seen together in AIHE lesions, HSP is more characterized by palpable ecchymotic (palpable purpura) lesions. While there is no visceral involvement in AIHE, involvement of other systems (gastrointestinal, kidney, etc.) is one of the findings of HSP. AIHE is usually seen in individuals aged 2 years, while HSP is more commonly seen in individuals between 3-10 years of age (1,17-19).

In differential diagnosis, it is necessary to distinguish AIHE from HSP, which is closely related to it; Kawasaki; Erythema multiforme; urticaria with hemorrhage (urticarial vasculitis); vasculitis caused by drugs; and conditions such as trauma and child abuse, which we would like to emphasize. For this purpose, as was done in our patient, fundus examination after a good anamnesis and trans-fontanel USG in individuals whose fontanels have not yet closed are the methods that can be used. Because purpuric rashes can occasionally occur after trauma, patients may recall trauma or child abuse. Purpuric rashes can be confused with AIHE, leukemia, and HSP rashes in children, especially by inexperienced physicians (20).

Child abuse is divided into 3 large groups: physical, sexual, and emotional abuse. Physical child abuse, which is related to our case, can be defined as the physical damage or injury to children under the age of 18 by their mother, father, or another person responsible for their care in a way that will harm their health. The possibility of physical abuse should be considered in every child presenting with an injury or signs of injury, such as purpuric rash. When this is suspected, a careful history should be taken, a physical examination should be performed, and routine radiological examinations and blood counts should be added to the examination (21).

In the evaluation made by taking the history, delay in bringing the child to the doctor, contradictory statements in the history, a history that does not match the physical results, recurrent and suspicious injuries, parents blaming the child or someone else for the injury, the child blaming the parents for the injury, a history of abuse in the parents, and the child appearing indifferent or overly anxious about the injury should suggest physical abuse. During physical examination, bruises in the calf/leg and genital area, soft tissue damage in different healing stages, special marks, such as hand/bite marks, numerous cigarette burns, liver or spleen rupture due to blunt abdominal trauma, cephalohematoma, subperiosteal hemorrhage, epiphyseal separation, metaphyseal fracture, radiological findings, such as periosteal calcification, retinal hemorrhage, eye damage, such as lens dislocation, multiple rib bone fractures, and ear damage with tympanic membrane rupture caused by traction, may be noted (22).

In some cases, upper respiratory tract symptoms, such as fever, cough, and dyspnea, were observed before lesions developed in patients with rhinovirus-based AIHE (23). Although upper respiratory tract infection was not identified in our patient, we believe that she may have been asymptomatic. In AIHE cases occurring after vaccination, lesions usually occur 2-3 weeks after vaccination (24–26). In our case, the fact that the lesions appeared very soon after vaccination and on the same day also distanced us from the etiology of vaccination. Once again, this is a case that perhaps shows that blaming vaccines as the cause of various diseases and disorders without being well-known and the reasons for vaccine hesitancy and rejection are not very accurate.

In conclusion, although AIHE can sometimes be confused with trauma, abuse, and hematological diseases in pediatric clinics, including emergency departments, it is a disease that can be easily diagnosed and has a benign course.

**Informed Consent:** Written consent was obtained from the participants.

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