CLINICAL IMAGE

Hemolacria, epistaxis, bloody otorrhea, hemoptysis, and hematuria in an 18-year-old man

Joanna Kosałka-Węgiel[1,2,3], Katarzyna Wawrzycka-Adamczyk[1,2], Aleksandra Matyja-Bednarczyk[1,4], Andżelika Siwiec[1,2], Anna Władarczyk[1,5], Stanisława Bazan-Socha[1,4]

1 2nd Department of Internal Medicine, Jagiellonian University Medical College, Kraków, Poland
2 Department of Rheumatology and Immunology, University Hospital in Krakow, Kraków, Poland
3 Department of Rheumatology and Balneology, Jagiellonian University Medical College, Kraków, Poland
4 Outpatient Clinic for the Immunological and Hypercoagulable Diseases, University Hospital, Kraków, Poland
5 Department of Intensive Care and Perioperative Medicine, Jagiellonian University Medical College, Kraków, Poland

An 18-year-old man, not previously treated chronically, was admitted to the 2nd Department of Internal Medicine of Jagiellonian University College in Kraków due to a 3-year history of recurrent hemolacria, epistaxis, bloody otorrhea, hemoptysis, and hematuria (FIGURE 1A and 1B). All his symptoms had been usually related to irritant exposure, such as indoor spaces with recently washed and disinfected floors, windows, or tables, as well as cold weather and cigarette smoke. Therefore, he spent most of his time at home avoiding public areas. He had no history of trauma. Clinical examination revealed diminished breath sounds on the right side and diffuse expiratory wheezing. The remainder of the physical examination was unremarkable.

The initial workup was notable for erythrocyturia. His complete blood count, peripheral blood smear, prothrombin time, activated partial thromboplastin time, fibrinogen level, serum tryptase concentration, platelet aggregation induced by adenosine diphosphate, collagen, ristocetin, epinephrine, and arachidonic acid were within reference ranges. Likewise, von Willebrand factor ristocetin cofactor activity, plasma levels of von Willebrand factor antigen, and coagulation factors II, V, VII, VIII, IX, X, XI were within reference ranges. Serologies for HIV, hepatotropic viruses, *Chlamydia pneumoniae* immunoglobulin (Ig) A and IgM, *Mycoplasma pneumoniae* IgM were negative. Blood tests revealed no signs of autoimmunity (antinuclear antibodies, antineutrophil cytoplasmic antibodies, rheumatoid factor, anticitrullinated protein antibodies, antitransglutaminase antibodies, and antiendomysial antibodies). Further immunologic tests (peripheral lymphocyte phenotype, levels of IgE [total and allergen specific], IgA, IgG [total with 1–4 subclasses], and IgM) were also within the reference ranges. A biopsy specimen of a new erythematous skin rash on the right forearm revealed no features typical

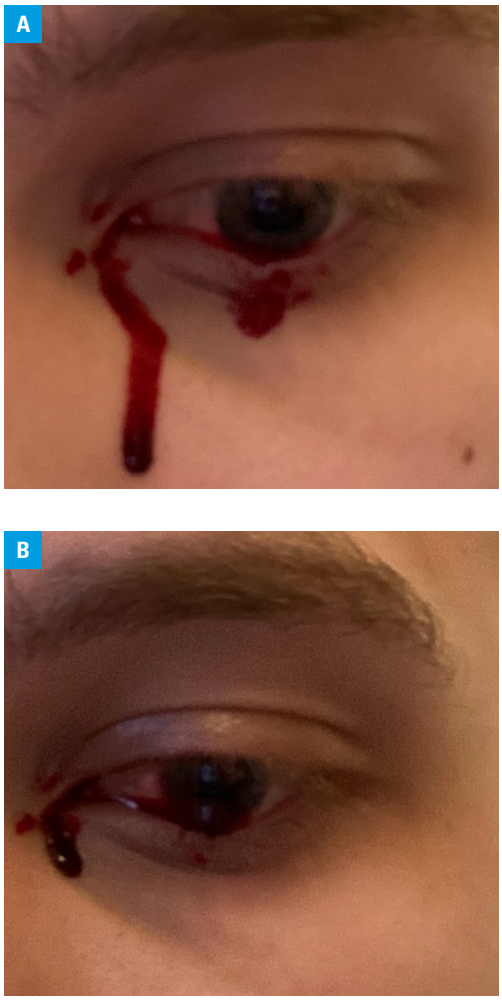

FIGURE 1 A, B – the left eye with hemolacria a few minutes after irritant product exposure

Correspondence to:
Joanna Kosałka-Węgiel, MD, PhD, Department of Rheumatology and Balneology, Jagiellonian University Medical College, ul. Jakubowskiego 2, 30-688 Kraków, Poland, phone: +48 12 400 31 10, email: joanna.kosalka@uj.edu.pl
Received: May 3, 2021.
Revision accepted: June 7, 2021.
Published online: June 8, 2021.
Pol Arch Intern Med. 2021; 131 (7-8): 735-736
doi:10.20452/pamw.16017

of mastocytosis. Paranasal sinus computed tomography and facial magnetic resonance imaging showed mild mucosa thickening of the left maxillary sinus, which was clinically irrelevant. Chest high-resolution computed tomography indicated no pathology; however, previous workup (performed 7 months earlier) showed an elevated eosinophil count (41% of cells) in bronchoalveolar lavage fluid. We excluded parasitic infections. Abdomen computed tomography showed mild hepatosplenomegaly, albeit without lymphadenopathy. During further hospitalization, an episode of massive hemolacria, epistaxis, bloody otorrhea, and hemoptysis with respiratory failure occurred after exposition to irritant cleaning products. We started dexamethasone intravenously, inhaled bronchodilators, and passive oxygen therapy with rapid clinical improvement. Finally, we diagnosed mucosal bleeding diathesis, likely related to eosinophilic tissue infiltration, and recommended local and systemic low-dose glucocorticoid therapy with loratadine. The patient's clinical response was good; however, he had a few episodes of hemolacria, bloody otorrhea, and hematuria in the first 2 months following discharge. During the next 40 months, he was asymptomatic, which is associated with an excellent clinical prognosis. Currently, he returned to normal life. However, we recommended avoiding solid inhaled irritants, such as paints or other chemicals, and performing a complete blood count and urinalysis every 6 months. The results have been within reference ranges so far. We believe that this is the first case report in Poland describing successful treatment of mucosal diathesis with antihistaminic drugs and glucocorticoids.

Hemolacria, which is characterized by the presence of blood in tears, can occur in the context of infection, inflammation, or trauma to the eye or surrounding structures, vascular tumors of the eye or surrounding structures, retrograde epistaxis, bleeding disorders resulting from systemic disease or medications, or psychogenic causes.[1,2] However, idiopathic hemolacria is rarely reported and requires a thorough examination and workup for diagnosis by exclusion.[2] It may be accompanied by hematohidrosis, bloody otorrhea, epistaxis, hematochezia, and hematuria.[2-4] Treatment options depend on the etiology.[1,5] Tissue eosinophilia, especially in the context of hemolacria and other systemic bleedings, should prompt the administration of antihistaminics and glucocorticoids to prevent severe or fatal progression of the disease.

ARTICLE INFORMATION

CONFLICT OF INTEREST None declared.

HOW TO CITE Kosałka-Węgiel J, Wawrzycka-Adamczyk K, Matyja-Bednarczyk A, et al. Hemolacria, epistaxis, bloody otorrhea, hemoptysis, and hematuria in an 18-year-old man. Pol Arch Intern Med. 2021; 131: 735-736. doi:10.20452/pamw.16017

REFERENCES

1 Di Maria A, Famà F. Hemolacria – crying blood. N Engl J Med. 2018; 379: 1766.

2 Sobol EK, Barmettler A. A case of idiopathic bilateral hemolacria in an 11-year-old girl. Ophthalmic Plast Reconstr Surg. 2017; 33: e98-e99.

3 Das D, Kumari P, Poddar A, et al. Bleeding to life: a case series of hematohidrosis and hemolacria. Indian J Pediatr. 2020; 87: 84.

4 Manzano G, Shantharam R, Webb E, et al. Case 2: hemolacria, hematochezia, and hematuria in an 11-month-old boy. Pediatr Rev. 2018; 39: 418-420.

5 Praveen BK, Vincent J. Hematidrosis and hemolacria: a case report. Indian J Pediatr. 2012; 79: 109-111.