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Case Report

Anaphylaxis followed by unilateral lung opacity and hypocomplementemia in a young female

Abstract

A 36-year-old woman was stung in the right wrist by a bee, suffered typical anaphylaxis, and was transferred to a local hospital. After a few hours, which corresponded to late-onset reaction, she developed shortness of breath and weakness and was transferred to the emergency department, where the diagnosis of anaphylaxis was confirmed. Serum complement levels, components C3 and C4, were undetectable. Flexible bronchoscopy excluded lung hemorrhage. She was on the ventilator for 4 days and was fully awake during that time. After the treatment, her improvement was rapid, and she fully recovered. Three weeks after she had been stung by a bee, skin prick tests to bee, wasp, and yellow jacket venom were done, and all tests came back negative as well as IgE and IgG4 antibodies to the same venoms. Coagulation factors and the complement, including C1q inhibitor, were normal. Occurrence of complement activation and consumption could point to the immune complexes as basis of pathophysiological mechanism. It remains unclear why such a reaction would involve only the right lung and why no detectable immune complexes were discovered. The clinical picture in the presented case resembled acute respiratory distress syndrome, but the exact nature of lung consolidation remains puzzling. The most likely explanation is a very rare case of airway obstruction. To our knowledge, this is the only reported case of anaphylaxis associated with undetectable serum complement levels. The potential role and diagnostic significance of hypocomplementemia in cases of anaphylaxis should be further investigated.

A young woman, a doctor by profession (36 years of age, 2 children, pediatrician on a postgraduate training at University Children's Hospital), was stung in the right wrist by a bee. She has suffered typical anaphylaxis (urticaria and angioedema, collapse, low heart rate, low blood pressure, vomiting, diarrhea) and was transferred by an ambulance to a local hospital. After a few hours, which corresponded to late-onset reaction, she developed shortness of breath and weakness and was transferred to the emergency department. Chest x-ray showed consolidation of the right lobe (Figure). She was hypoxemic, and blood pressure was low (85/65 mm Hg). Serum tryptase was highly elevated (76 ng/L), confirming the diagnosis of anaphylaxis. Blood gasses were compatible with diagnosis of acute respiratory distress syndrome (ARDS) (Pao_2/fraction of inspired oxygen ratio, 167), although lung opacity was unilateral. Her erythrocyte sedimentation rate, white blood cell count, and C-reactive protein were normal. Bacteriology was negative, and no signs of pneumonia were detected. Flexible bronchoscopy excluded lung hemorrhage, and tracheobronchial tree was completely normal. Serum complement levels, components C3 and C4, were undetectable. Properdin was normal. She was on the ventilator for 4 days intermittent mandatory ventilation and during that time, she was fully awake. She was being given intravenous steroids, antihistamines, and antibiotics and fluids. Her improvement was fast, and she was weaned from ventilator after 4 days. Her recovery was full, chest X-ray was normal, and lung function tests showed normal lung function. Three weeks after she had been stung by a bee, skin prick tests to bee, wasp, and yellow jacket venom were done, and all tests came back negative. IgE and IgG4 antibodies to the same venoms were also negative. Coagulation factors were normal. The complement was also normal, including C1q inhibitor.

Unusual or unexpected reactions occur in a temporal relationship after insect stings [1]. These reactions are very infrequent, differ from typical allergic symptoms, and often do not have an apparent cause. Therefore, they are classified by the organ system affected (cardiac, renal, or neurologic), the local or systemic manifestations, and the timing of the symptoms. In this case, occurrence of symptoms clearly indicated biphasic reaction [2]. Occurrence of complement activation and consumption could point to the immune complexes as basis of pathophysiological mechanism. It is not clear why would such reaction involve only the right lung and why no detectable immune complexes were discovered during immunology workup. The

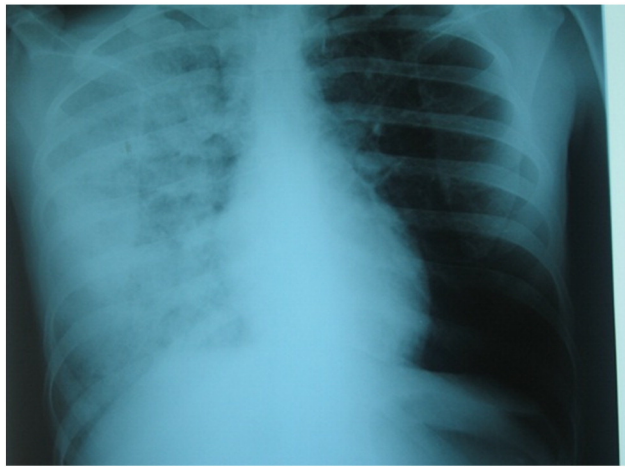


Figure. Posteroanterior chest radiographs showing unilateral lung opacity in a 36-year-old woman with anaphylaxis.

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presented case clearly differs from previously described cases of alveolar hemorrhage because no alveolar hemorrhage could have been demonstrated (bronchoalveolar lavage cytology showed no red blood cells, and induced sputum 2 weeks later showed no evidence for iron laden macrophages) [3,4]. The patients described in those 2 cases reacted to bee sting, which suggests that the substance that started reaction is common to bee venom.

The clinical picture in the presented case resembled ARDS, but the exact nature of lung consolidation remains puzzling. By definition, lung opacity in ARDS is bilateral. Unilateral lung edema occurs during surgery (positional), after lung reexpansion and several other conditions, none of which was present in our patient [5]. The most likely explanation is a very rare case of airway obstruction. To our knowledge, this is the only reported case of anaphylaxis associated with undetectable serum complement levels. The potential role as well as diagnostic significance of hypocomplementemia in cases of anaphylaxis should be further investigated.

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References

[1] Reisman RE. Unusual reactions to insect stings. Curr Opin Allergy Clin Immunol 2009;5(4):355–8.
[2] Ellis AK, Day JH. Incidence and characteristics of biphasic anaphylaxis: a prospective evaluation of 103 patients. Ann Allergy Asthma Immunol 2007;98(1):64–9.
[3] Mukhopadhyay A, Fong KF, Lim TK. Diffuse alveolar haemorrhage: a rare reaction to insect sting. Respirology 2002;7(2):157–9.
[4] Rueff F, Przybilla B, Bilo MB, Muller U, Scheipl F, Aberer W, et al. Predictors of severe systemic anaphylactic reactions in patients with Hymenoptera venom allergy: importance of baseline serum tryptase—a study of the European Academy of Allergology and Clinical Immunology Interest Group on Insect Venom Hypersensitivity. J Allergy Clin Immunol 2009;124(5):1047–54.
[5] Slama K, Gesch M, Bock JC, Pietschmann SM, Schaffartzik W, Pison U. Unilateral lung edema: effects on pulmonary gas exchange, hemodynamics, and pulmonary perfusion distribution. J Appl Physiol 2000;89(4):1513–21.

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