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Fatal anaphylaxis after a yellow jacket sting, despite venom immunotherapy, in two patients with mastocytosis

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VIT Venom-specific immunotherapy

Death after a yellow jacket sting in otherwise healthy individuals receiving adequate immediate therapy is an uncommon occurrence.¹ Mastocytosis is overrepresented in patients first seen with anaphylaxis after insect stings, but it remains a rare disorder, even in this patient group.²

This report describes two patients seen at our center with confirmed mastocytosis who died after a yellow jacket field sting, despite optimal treatment. Mastocytosis may be a risk factor for a severe or fatal reaction after Hymenoptera stings in some patients with this disease.

Case Reports

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Patient 1

A 44-year-old woman came to our clinic because of a severe systemic reaction after a yellow jacket sting in 1980. The skin test response to yellow jacket venom was positive at 1 µg/ml. Venom-specific immunotherapy (VIT) with yellow jacket venom was started and discontinued after 5 years without side effects or intercurrent stings.

In 1989 she returned to our clinic for treatment of skin lesions and was diagnosed as having urticaria pigmentosa by means of skin biopsy. She had no other symptoms of mastocytosis. The diagnosis of systemic mastocytosis was confirmed by the finding of elevated baseline urinary methylhistamine (163 to 253 µmol/mol creatinine; upper limit of normal, 150 µmol/mol creatinine) and a positive bone marrow biopsy result. She required no medication but was advised to carry an Epipen (ALK, Denmark).

In July 1994, she was stung by a yellow jacket. She did not use her Epipen. A general practitioner reached her within 2 minutes. The patient had collapsed, and the physician administered 1 mg of epinephrine and 2 mg of clemastine, both intramuscularly.

Paramedics arrived within 12 minutes and found the patient unconscious with depressed respirations, no measurable blood pressure, and an initially weak and rapid pulse, which became absent shortly thereafter. Resuscitation was started. Epinephrine, 1 mg, was administered intravenously. Her pulse was restored with a frequency of 115 beats/min, and spontaneous respirations returned.

Shortly thereafter, she went into cardiac arrest. Resuscitation was restarted; and the patient was intubated, stabilized, and transported to a local hospital where she arrived 45 minutes after her collapse.

On arrival at the hospital, the patient was unconscious with sluggishly reacting pupils. One hour later she had a convulsion, and postischemic cerebral edema subsequently developed, as confirmed by computed tomographic scan. On the fifth day the diagnosis of cerebral death was established.

Patient 2

A 43-year-old woman experienced a severe systemic reaction in 1989, after a probable yellow jacket sting. The skin test response to yellow jacket venom was subsequently positive at 0.1 µg/ml, and the RAST result was weakly positive (class 1, 0.43 PRU/ml).

VIT was started, but before the maintenance dose was reached, she had a systemic reaction after a yellow jacket field sting. Immunotherapy was subsequently continued.

Two and a half years into VIT, she experienced a systemic reaction 30 minutes after a maintenance dose injection. One month later, she was re-evaluated and again underwent skin testing, which again resulted in a systemic reaction to 0.01 mg/ml. VIT was discontinued and the patient was referred to our hospital. The diagnosis of mastocytosis was established by elevated baseline urinary methylhistamine levels (201 to 253 µmol/mol creatinine), urticaria pigmentosa (upper arms and upper legs) as confirmed by skin biopsy, and a positive bone marrow biopsy result. She had no symptoms of mastocytosis and required no medication. She continued to carry an EpiPen.

In July 1994, 15 months after stopping VIT, she was stung by a yellow jacket. Although she used the EpiPen within minutes, she lost consciousness. An ambulance arrived 10 minutes later, and paramedics found the patient with no detectable pulse and gasping respirations. Resuscitation was started; and epinephrine, clemastine, and prednisolone were given intravenously. She was intubated and transported to a local hospital, where she was found to be comatose and cyanotic with undetectable pulse and blood pressure and fixed, dilated pupils. Resuscitation was continued for 1 hour, during which she received several intravenous administrations of epinephrine, as well as other medications, including clemastine and dopamine. Cardiac monitoring showed complete A-V dissociation with progressively widening QRS complexes and persistent electromechanical dissociation. After assessment by a neurologist, she was pronounced dead, and resuscitation was terminated.

Discussion

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Mastocytosis may manifest with recurrent episodes of severe anaphylaxis, including profound hypotension, but fatal anaphylaxis is rare.³ Anaphylactic shock after a Hymenoptera sting can be an initial symptom of systemic mastocytosis,² even in the absence of specific IgE.

Fatal reactions after Hymenoptera stings are rare, with a prevalence of 0.09 to 0.45 deaths per year per million.¹ Possible risk factors include the presence of atherosclerosis, coronary artery disease, and/or chronic obstructive pulmonary disease in older patients.¹ Both of the patients described here were relatively young, had no identifiable risk factors, and had only an average disease load as determined by urinary methylhistamine levels.⁴ Both received adequate support after their field sting, including prompt administration of adequate doses of epinephrine, which is probably the most important therapeutic determinant of a good outcome.⁵

VIT is an effective treatment for the prevention of anaphylaxis in patients with previous systemic reactions after Hymenoptera stings. Although VIT has been used to treat patients with mastocytosis and anaphylaxis after Hymenoptera stings, the efficacy of this treatment in this group of patients

has not been well documented. Engler and Davis⁶ described one such patient who successfully underwent rush VIT and tolerated a field sting without experiencing a systemic reaction. Both our patients had VIT: patient 1 for 5 years, which is considered to be sufficient for the long-term prevention of anaphylaxis, and patient 2 for at least 2 years. To our knowledge, patient 1 represents the first reported case of fatal anaphylaxis, despite optimal VIT.

These cases demonstrate that not all patients with mastocytosis and anaphylactic sensitivity to Hymenoptera venom have a good prognosis. Disease load did not seem to be an important prognostic factor in our two patients, and other indicators of a poor prognosis were not evident. Neither patient was highly sensitized, and the post mortem yellow jacket venom RAST result was negative in patient 1 in whom this was done.

Although deaths after Hymenoptera stings remain rare, their impact on the community is disproportionately great. Our recent experience reaffirms our belief that screening patients first seen with Hymenoptera anaphylaxis for mastocytosis by means of an appropriate laboratory test, such as measurement of urinary methylhistamine, is informative and useful. The diagnosis contributed to our understanding of the clinical courses of these two patients, whose deaths would have otherwise been regarded as straightforward therapeutic failures. In view of the lack of efficacy of conventional acute and preventive treatment in some patients with mastocytosis, individuals with this disease should probably be advised to practice maximal sting avoidance.

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