

Factitious Hymenoptera allergic emergency: A report of a new variant of Munchausen's syndrome

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We report the case of a young woman with three hospitalizations and six emergency room visits for treatment of bronchospasm and angioedema first thought to be the result of Hymenoptera venom allergic reactions. Because several features of her illness were not consistent with the usual course of Hymenoptera anaphylaxis and because of a known history of anaphylactic-type reactions following aspirin ingestion, a presumptive diagnosis of factitious anaphylaxis was considered, the medical staff was alerted to evaluate this possibility, and the diagnosis was established. Serologic testing supported the diagnosis. This variant of Munchausen's syndrome is a third type described as an allergic emergency. The others are peanut anaphylaxis and self-induced laryngeal stridor.

Numerous cases of factitious medical and surgical emergencies have appeared in the medical literature with several general reviews.¹⁻³ Factitious diseases that are allergic emergencies or simulate such emergencies have been rarely reported.⁴⁻⁶ Systemic IgE-mediated reactions or diseases that mimic them constitute some of the most acute of medical emergencies. A young woman who appeared to react to Hymenoptera venom was evaluated and after investigation a diagnosis of Munchausen's syndrome was made. Because of similarities noted between this patient and other patients seen previously in this service with factitious allergic emergencies, we present a review of our experience.

CASE REPORT

A 23-yr-old woman reported to the emergency room of Northwestern Memorial Hospital stating she had been stung by a "bee" while driving her automobile 24 hr earlier. She reported mild papules at the site of the presumed sting on

her right hand and localized swelling about the eyes and mouth. There were no other abnormalities noted on physical examination and the patient was treated with diphenhydramine and released from the emergency room with a presumed diagnosis of delayed insect sting allergy. She returned to the emergency room 4 hr later in severe respiratory distress. No exacerbating event for this change of clinical status was evident. She had a respiratory rate of 35 and by auscultation distant shallow breath sounds were noted with scattered expiratory rhonchi and rales. The pulse rate was 120. Clinical evaluation also suggested inspiratory stridor but indirect laryngoscopy revealed no edema of the vocal cords or upper airway tissues. The patient was treated with subcutaneous epinephrine and intravenous hydrocortisone hemisuccinate and aminophylline with resolution of her symptoms within 1 day following hospital admission.

An allergy consultation was obtained. The patient insisted her symptoms were caused by the previous insect sting and insisted there could be no other possible explanation. She stated her mother and brother had suffered similar "allergic reactions" to honeybees around her home in the spring and summer. Her past medical history was significant for trauma to her lower back following an automobile accident and for which an extensive medical work-up was required because of chronic pain. No pathology other than soft tissue injury was found. Her social history was also significant. She admitted she had not spoken with her parents for a period of years because of their cruelty to her and she did not trust them. In addition she described a history of aspirin and codeine intolerance dating from childhood with urticaria and bronchospasm after ingestion of either drug. She claimed careful avoidance of these medications. Her

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TABLE I. Skin-testing results with intradermal Hymenoptera venoms (Pharmacia)

	Honeybee	Wasp	Yellow jacket	Yellow-faced hornet	White-faced hornet
0.1 μ g/ml	0	0	0	0	0
1.0 μ g/ml	0	0	+	+	+

work involved research with patients suffering mental depression at a local medical institution. She admitted anxiety and depression over the recent loss of several companions including an intimate partner.

The patient remained asymptomatic during hospitalization until she returned to her room following routine pelvic examination at which time she again suffered the acute onset of severe bronchospasm. The symptoms responded to treatment with epinephrine and aminophylline followed by 40 mg of prednisone daily. During her hospitalization the following tests and evaluations were normal: complete blood count, differential leukocyte count, SMA-12 chemistries, electrolytes, urinalysis, chest x-ray film, electrocardiogram, total complement, C3, C4, C1 esterase inhibitor and total serum immunoglobulins.

Because these acute episodes of angioedema and bronchospasm occurred over a period of 24 to 72 hr following presumed insect sting, it was concluded by the allergy service that Hymenoptera sensitivity should be excluded. Skin tests were performed following hospital discharge and the results are shown in Table I. Additional immunologic evaluations were made in the allergy laboratory of Northwestern University and by Dr. John Yunginger of the Mayo Clinic.

Although our patient left the outpatient clinic following skin testing in good health, she returned the following day to the clinic in severe respiratory distress. She complained that wheezing and a feeling of chest tightness followed her skin testing with venoms and this was accompanied by swelling at the injection sites. On physical examination she was noted to have a respiratory rate of 30, pulse of 120, and diffuse expiratory rhonchi. There was no evidence of any local reaction. She responded well to epinephrine and was released from the clinic with instructions to take 40 mg prednisone daily for 1 wk. However, the next day she returned to the emergency room, again in obvious distress. She insisted her dyspnea was caused by "reactions" to her previous venom skin testing. The patient was admitted to the hospital and although the diagnosis of self-induced bronchospasm by aspirin or codeine was suspected, salicylate in the blood could not be detected at the time of analysis. Within 24 hr of admission, she was again asymptomatic and was discharged. The following weekend she called the emergency allergy service to report an episode of wheezing that occurred on a local beach following stings by mosquitos and other "flying insects." A week later she reported to the emergency room in severe respiratory distress and once again received intravenous treatment for bronchospasm. Factitious allergic disease was again considered and a physician who had treated her since her initial referral to the

TABLE II. Polystyrene tube binding of IgE against phospholipase A

Test serum samples	¹²⁵ I-phospholipase A (ng) bound to tube
Pooled honeybee worker sera	10.1
Pooled vespid allergic patients' sera	1.2
Normal control patients	0.1
Patient with suspected Hymenoptera allergy	0.1

allergy service was called to speak with her. The patient related her numerous personal problems previously reported and stated that although she suffered only mild attacks of wheezing at home, that personal insecurities caused her to react to mild dyspnea with great anxiety. When assured the staff would offer support regardless of any medical or psychiatric problems she might suffer, the patient admitted she had induced the episodes with aspirin and at this time gave the medical staff a bottle of aspirin concealed in her purse.

Our patient was referred for psychiatric evaluation and was subsequently admitted to the psychiatric service three times for treatment of severe depression. During one of these admissions, she again suffered an episode of bronchospasm requiring treatment. Salicylate level drawn during this attack was 4 mg/100 ml.

MATERIALS AND METHODS

Measurement of total serum blocking antibody against phospholipase A

Blood specimens were drawn from the patient on three occasions. The sera were analyzed for antibody against honeybee phospholipase A by a modification of the ammonium sulfate technique of Lidd and Farr.⁷⁻⁹ Briefly, honeybee phospholipase A (Sigma) was radiolabeled with ¹²⁵I.¹⁰ Aliquots of 0.1 ml of sera were incubated with 10, 100, and 1,000 ng of ¹²⁵I-phospholipase A. After appropriate incubation, the antibody-bound phospholipase A was separated from free phospholipase A by precipitation with 40% saturated ammonium sulfate. The nanograms of phospholipase A bound by 1 ml serum in antigen excess were calculated.

Measurement of serum IgE antibody against phospholipase A

The sera were analyzed for IgE against phospholipase A by a modification of the polystyrene tube radioimmunoassay

TABLE III. Percent of the total counts per minute of radiolabeled anti-IgE bound to microcrystalline cellulose

	Honeybee	Wasp	Yellow jacket	Yellow-faced hornet	White-faced hornet
Negative control group	0.48	0.27	0.34	0.29	0.38
Positive control group	9.14	3.94	6.26	3.27	4.38
Patient with suspected Hymenoptera allergy	0.92	0.60	0.52	0.70	0.46

technique of Zeiss et al.¹¹ The results are reported as nanograms of phospholipase A bound by IgE in 1 ml of serum.

Measurement of serum IgE against vespid antigens by RAST

Blood specimens were analyzed for the presence of IgE against a panel of Hymenoptera venoms by Dr. Yunginger. Briefly, Hymenoptera venoms were covalently coupled to microcrystalline cellulose particles and after initial incubation with sera, the solid-phase allergens with attached IgE were washed and radiolabeled anti-IgE added in the second stage.¹²

RESULTS

Serum blocking antibody against phospholipase A

Fig. 1 shows the percent ¹²⁵I-phospholipase A bound to globulin plotted on the y axis of semilog paper and the nanograms cold phospholipase A added to the tube plotted on the x axis. The nanograms of phospholipase A bound at the 20% binding point were multiplied by the dilution factor. The result is the nanograms of phospholipase A bound by 1 ml of serum in antigen excess. Results are shown for pooled sera obtained from honeybee- and vespid-allergic patients (courtesy of Dr. Yunginger), for normal control patients, and for patient 1 with suspected Hymenoptera allergy.

Serum IgE against phospholipase A

Table II shows the results of polystyrene tube radioimmunoassay for IgE in sera against phospholipase A. Results are expressed as nanograms of phospholipase A bound per 1 ml of serum and are shown for honeybee workers, vespid-allergic patients, normal controls, and for patient 1 with suspected Hymenoptera allergy.

Serum IgE against Hymenoptera venom antigens by RAST

Results are shown in Table III and are expressed as the percent of the total counts per minute of

radiolabeled anti-IgE bound to microcrystalline cellulose and are proportional to serum IgE against each allergen.

DISCUSSION

We conclude that the case presented demonstrated that the patient who first was considered to have possible Hymenoptera anaphylaxis did not have such a sensitivity based on her clinical course, skin tests, and laboratory studies. Further, a self-induced systemic reaction to aspirin was suspected, admitted to by the patient, and confirmed on one occasion by blood salicylate level. The multiple admissions and emergency-room visits this patient required for self-induced reactions permit classification of her problem as Münchhausen's or a hospital-addiction syndrome. Two other variants of this syndrome have been reported and are summarized here.^{4, 5}

Case 2

Four years ago we reported the history of a young woman presenting with complaints judged to be repeated factitious allergic emergencies. A 20-yr-old, unmarried white female student nurse was referred to the allergy service following hospitalization for an episode of acute urticaria, rhinitis, angioedema, and asthma. The patient, during interview, admitted an earlier history of severe generalized reactions similar to the above complaints after ingestion of nuts. She denied any known recent ingestion of nuts but over the next 4 wk suffered three episodes of angioedema and asthma, the last of which required hospitalization. On a later ambulatory clinic visit, another acute reaction occurred and after appropriate therapy, the patient was removed to an observation area and her belongings were examined. The patient's purse contained a plastic bag with cracked Brazil nuts and filberts. Urgent psychiatric consultation and confrontation of the patient with the evidence of nuts led to the admission of nut ingestion on that occasion.

TABLE IV. Summary of three patients with Munchausen's syndrome

Patient	Personal profile	Factitious emergency	Means of induction
1	Single female, aged 23 yr; worked in health-related field with physicians; recent loss of several friends and intimate associate; two psychiatric admissions for suicide gestures; claimed mother was alcoholic and father often beat her; psychiatric consult judged patient to suffer from "severe depressive neurosis"	Bronchospasm, urticaria, and angioedema requiring 6 emergency room visits and 3 hospitalizations blamed on allergic reactions to stinging insects	Presumed salicylate ingestion documented by positive blood salicylate level on one occasion and aspirin found in purse
2	Single female, aged 20 yr; employed as nurse; recent loss of fiancé; admitted suicide gesture as reason for nut ingestion; refused psychiatric evaluation	Angioedema and urticaria simulating idiopathic or allergic anaphylaxis documented on 3 clinic visits and requiring hospitalization	Presumed nut ingestion documented by finding nuts in the purse of patient during an attack of angioedema in the clinic
3	Separated female, aged 32 yr; unemployed; never left alone by her mother or grandmother because of concern over her attacks; refused psychiatric evaluation	Presumed laryngeal edema requiring 13 emergency admissions to local hospitals, each time presenting with symptoms of stridor	Mimicry thought to be single cause of induction of symptoms; medical work-up revealed no abnormalities of airways or chest

Case 3

Five years ago a 32-yr-old married but separated woman appeared in the emergency room in acute respiratory distress. The symptoms were tachypnea (45 breaths per minute) and marked inspiratory stridor. A diagnosis of laryngeal obstruction was made and immediate therapy with epinephrine, antihistamines, and intravenous hydrocortisone was instituted. Examination of the vocal cords by indirect laryngoscopy showed no evidence of obstruction. When detailed history could be obtained, it was learned this patient had required at least 15 emergency admissions to several hospitals for similar symptoms and signs. No precipitating events could be ascertained. There was no history of respiratory or atopic disease. Because of these attacks, she had stopped working and was never left alone by her mother because of concern over her attacks. Laboratory functions found to be normal were complete blood count, SMA-12 chemistries, electrolytes, urinalysis, chest x-ray film, electrocardiogram, serum complement, C3, C4, C1 esterase inhibitor, quantitative immunoglobulins, and pulmonary function testing including inspiratory flow parameters. Lateral x-ray films of the neck were normal. Following admission, all clinical signs of laryngeal obstruction abated within 8 hr. Although tentative diagnosis included foreign body aspiration, retained secretions, laryngeal edema, and tumor of a large airway, no evidence for these could be found. During a second attack, immediate direct laryngoscopy again

demonstrated the vocal cords were normal. The patient was encouraged to cough on several occasions and, thus distracted by this activity, she was observed to take normal breaths immediately before and after coughing, showing that her stridor was nonorganic.

We have now reported three cases of factitious allergic emergencies. As in other reviews of Munchausen's syndrome, we have noted similarities between each of these patients. A tabulation is made in Table IV. General aspects and the psychopathology of Munchausen's syndrome were reviewed by Ireland et al.¹ There appears to be a similarity among the cases of Munchausen's syndrome that have been reported in the literature since 1951. It appears predominantly in countries where hospitalization is free. It primarily involves young middle-aged adults with onset occasionally traced to teenage years. What becomes of these patients in later years is poorly documented. Some might die of self-induced diseases or as they grow older lose the vitality necessary to maintain their hospital addiction. All of the patients reported appear to be socially isolated without family ties or close personal relationships of any kind and reliable family and social histories are difficult to obtain. The most remarkable characteristic shared by the group is their marked ability for self-destruction.¹³ Prognosis for Munchausen's syndrome is thought to be poor but an attempt should be made to establish a rational therapeutic program. The first step is a tolerant and sympathetic attitude. Arrangements should be made for

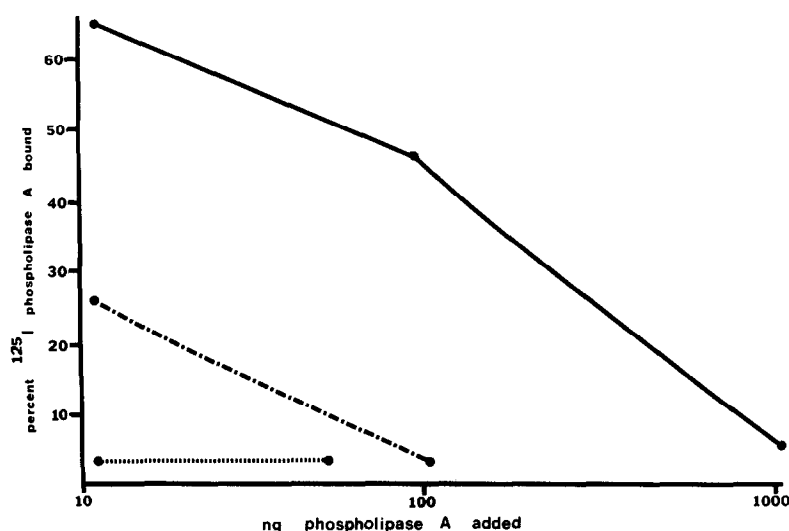


FIG. 1. Ammonium sulfate precipitation of radiolabeled phospholipase A bound to immunoglobulin. Phospholipase A (nanograms) bound at 20% binding point was multiplied by the dilution factor. ■■■, Binding for honeybee workers' serum pool. Phospholipase A bound per milliliter serum was calculated as 900 ng. ---, Binding for vespid-sensitive patients. Phospholipase A bound per milliliter serum was calculated as 40 ng. □□□, Binding for patient's serum and for normal controls.

long-term supervision and psychotherapy by one or several physicians interested in the patient.

Two of our patients worked in medically related fields and all were either single or separated. A significant factor in their illnesses appeared to be isolation from strong family support. All three patients were quite resistant to psychiatric referral and our patient with nut-mediated allergy stopped her appointments. Our patient presenting with signs of allergic emergency following Hymenoptera stings, which we believe is factitious, still insists she is allergic to the stings of honeybees despite our evidence against this.

The diagnosis of factitious allergic emergency was suspected by history and confirmed by observation and confrontation in each of these three patients. Although the clinical history of our patient reported here was atypical of Hymenoptera allergy, extensive immunologic testing was performed during her evaluation by our service (Fig. 1 and Tables I, II, and III). Based on her complete clinical history, the results of her skin tests were interpreted as nonspecific irritative reactions to high concentrations of venoms used. A venom concentration of 1,000 ng/ml can induce a nonspecific reaction in normal skin.¹⁴ Results of radioallergosorbent test (RAST) against a panel of venoms performed by Dr. Yunginger supported this conclusion. However, a number of patients have been reported who suffered anaphylaxis following insect sting in whom venom-specific IgE could not be dem-

onstrated by RAST or skin testing.¹⁵ Whether these patients have undetectable levels of IgE antibodies or have their reactions because of other immune mechanisms is unclear. Analysis of IgE and total serum binding of honeybee phospholipase A was performed in our laboratory because the patient initially identified honeybee as the stinging insect responsible for her symptoms. Ninety percent of the total IgE antibody against venom can be removed from sera of honeybee-sensitive patients by absorption with phospholipase A.¹⁶ Although the phospholipase found in Apoidea venom is immunologically distinct from that of the venoms of the vespid groups, our pooled sera from vespid-sensitive patients contained small amounts of IgE specific for honeybee phospholipase. We assume that certain vespid-sensitive individuals have also been stung in the past by honeybees, leading to an immune response without clinical manifestations.

After review of our patient's clinical course we conclude she simulated Hymenoptera-allergic emergencies on several occasions by ingestion of aspirin. Although currently available immunologic techniques supported this analysis, laboratory determinations cannot discriminate between all allergic and normal individuals.

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