


Factitious Disorder Masquerading as a Life-Threatening Anaphylaxis

Journal of Investigative Medicine High Impact Case Reports
Volume 9: 1–4
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DOI: 10.1177/23247096211006248
journals.sagepub.com/home/hic


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Abstract

Factitious disorder is a psychiatric disorder in which sufferers intentionally fabricate physical or psychological symptoms in order to assume the role of the patient, without any obvious gain. We present a case of a 23-year-old female with chronic urticaria who presented with dyspnea, dysphasia, mild generalized erythema, abdominal cramps, and headache. She was tachypneic and hypotensive. This was her third admission with similar symptoms within the last 7 months. Tryptase, complement, anti-SM/RNP, Sjogren, Scl-70, C3, and C4 were negative. Computed tomography-guided bone marrow biopsy showed no mast cells. Flow cytometry did not show any immunophenotypic reaction. Other possible differentials including pregnancy, autoimmune disorders, and infections including hepatitis, thyroid disorder, and age-related malignancies were ruled out. After a thorough review, malingering disorder was ruled out, but we noticed the patient's intent of assuming a sick role. Later, the patient was diagnosed with major depressive disorder. Factitious anaphylaxis can present with multiple presentations including a life-threatening condition that mimics true anaphylaxis. A better approach would be thorough clinical evaluation and early multidisciplinary involvement. This case highlights the importance of further evidence-based studies in factitious disorder to decrease the disease burden and reduce the health care cost.

Keywords

factitious anaphylaxis, factitious disorder, anaphylaxis, major depressive disorder

Introduction

Factitious disorder (FD) is a psychiatric disorder in which sufferers intentionally fabricate physical or psychological symptoms in order to assume the role of the patient, without any obvious gain (American Psychiatric Association's *Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition*). Being a young adult, female gender, having an unmarried/divorced status, experiencing social isolation or family disruption at early age, being a health care worker, and having access to free hospitalization are reported as risk factors.^{1–5} Factitious anaphylaxis can present as a life-threatening condition, mimicking a true anaphylaxis that requires immediate medical attention.^{2,4,6} To our knowledge, only a few cases of factitious anaphylaxis have been reported.^{1–7} In this article, we present a case of a 23-year-old female with multiple emergency department (ED) visits and hospitalizations for episodes of anaphylactic symptoms with inconclusive laboratory studies, allergy testing, and imaging.

Case Presentation

A 23-year-old female with possible chronic urticaria presented to the ED due to difficulty breathing, difficulty swallowing,

mild generalized erythema, warm sensation, nausea, abdominal cramps, and headache for 1 day. The patient stated that she had chest tightness with feeling that her throat was closing. The patient also complained of having a burning sensation and some blisters after showering with lukewarm water. Her symptoms were relieved shortly after using an auto-injector epinephrine pen. In the ED, she was saturating at 100% on room air, tachypneic with respiratory rate of 31 breaths per minute, temperature of 37°C, heart rate of 72 beats per minute, and blood pressure of 134/89 mm Hg. Physical examination was significant for mild erythema over the face and chest with no hives or obvious wheals. Bilateral lung fields were clear to auscultation with no wheeze or stridor and with no use of accessory muscle. Neither dysphonia nor angioedema of lips and tongue were noted. On throat examination,

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Received February 26, 2021. Revised February 26, 2021. Accepted March 6, 2021.

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no signs of pharyngeal wall erythema or swelling was noted. Arterial blood gas showed pH of 7.39, PCO_2 of 35 and PO_2 of 71 on room air. Patient was admitted for further evaluation.

We found that the patient has had a pet cat for the past 4 years with no known allergic reaction. Home medications included cetirizine 10 mg, famotidine 40 mg, prednisone 20 mg twice a day, and epinephrine auto-injector pen. She denied any new medication use. On further evaluation, it was found that this was her third visit to our ED with similar symptoms within the last 7 months. During her first episode she had difficulty in breathing, which resolved after receiving intramuscular epinephrine. The second episode was 1 month ago when she presented with hives, angioedema of the eyes and lips, and difficulty breathing. Symptoms resolved after receiving IM (intramuscular) epinephrine. Later that day, her oxygen saturation dropped, which resolved with IV (intravenous) diphenhydramine and IM epinephrine. The following day, the patient was admitted to the intensive care unit (ICU) after she developed worsening hives without wheals. In the ICU, she was treated with epinephrine drip for 24 hours. She was discharged on oral cetirizine, famotidine, and prednisone and had no further episodes until this presentation.

During the current hospital stay, the patient had multiple episodes of allergic reactions to almost all oral intake, including food. Allergic reactions included subjective findings of dyspnea, dysphagia, flushing, nausea, and occasional vomiting and objective findings of sinus tachycardia in the range of 120 to 140 beats per minute with oxygen saturation of 100% on room air and no signs of respiratory distress. All episodes resolved with IV diphenhydramine 25 mg and IM epinephrine 0.3 mg. She also received a 9-day course of hydrocortisone 50 mg every 8 hours. Allergy specialist was consulted. Laboratory works included normal complete blood cell count without eosinophilia, normal serum immunoglobulin E level, negative tryptase, normal level of thyroid-stimulating immunoglobulin, anti-SM/RNP, Sjogren's, Scl-70, C3, and C4 levels. Blood, urine, and respiratory cultures were negative, so infection was ruled out. The hepatitis panel was negative. Radioallergosorbent test (RAST) indicated a negative response for beef, chicken, lamb, and pork even though the patient had an allergic reaction to all the food products. The patient underwent computed tomography-guided bone marrow biopsy, which showed no mast cells activity. Flow cytometry did not show any immunophenotypic reaction (Table 1).

In the next few days the patient had no episodes of allergic reaction. Multiple tests including repeat tryptase, workup to rule out carcinoid reaction, and pheochromocytoma were all negative. Psychiatry was consulted and the patient was discharged home with a diagnosis of somatoform disorder. Later, it was found that the patient had a complex history of family disruption and was diagnosed with major depressive disorder. On follow-up, it was found out that the patient

received electroconvulsive therapy in an inpatient psychiatric facility. She is currently on antidepressant with resolution of her allergic symptoms.

Discussion

Self-imposed FDs have multiple presentations. One of the rare presentations can be anaphylaxis. It is considered to be a specific subtype of somatoform disorder.¹

Our case is a young female presenting with multiple episodes of anaphylaxis with negative laboratory work, allergic testing, and bone marrow biopsy who was later diagnosed to have somatoform disorder and major depressive disorder. Multiple differentials for her presentation were considered. Chronic spontaneous urticaria also known as chronic idiopathic urticaria (CIU) is defined as a nearly daily occurrence of wheals with or without angioedema for more than 6 weeks.^{8,9} But our patient's symptoms were intermittent and she never exhibited obvious wheals. IgG auto-antibody against IgE or high-affinity IgE receptor (Fc ϵ RI) is found in approximately 40% of CIU cases, but multiple studies showed the presence of these antibodies even in healthy individuals and in individuals with other autoimmune disorder. This suggests that these antibodies are not unique to CIU.¹⁰⁻¹² Due to low specificity and inaccessibility of these tests in the setting of low suspicion of CIU, these antibodies were not tested in our case. The normal tryptase level and normal bone marrow biopsy in our case refute mast cell activity, which in turn rules out true anaphylaxis.¹ There was no suspicion of aspirin hypersensitivity as our patient tolerated multiple doses of oral and IV nonsteroidal anti-inflammatory drugs without any allergic reactions. There was no known history of any respiratory disorders including asthma and rhino sinusitis, physical examination revealed no nasal polyps, and laboratory tests ruled out eosinophilia, which can be associated with aspirin hypersensitivity. Serum IgE level was normal, RAST IgE for various foods were negative ruling out food allergies. Other possible differentials including pregnancy, autoimmune disorders, and infections including hepatitis, thyroid disorder, and age-related malignancies were ruled out.

After reviewing the clinical presentations and investigations, the authors ruled out the organic causes of anaphylaxis. We could not find any evidence of external gain, which rules out malingering disorder but noticed the patient's intent of assuming a sick role. In any atypical and refractory presentation of anaphylaxis with normal level of C1 inhibitor, one should consider FD.¹ Complete workup and management of the factitious anaphylaxis is often challenging given the nature of the clinical presentation and diagnostic uncertainty. In fact, the gain of attention during each intervention and hospitalization may even worsen the condition by reinforcing the patient's behavior of seeking further medical attention.⁶ One of the studies in Northwestern University's Division of Allergy and Immunology involving 350 patients calculated

Table 1. Laboratory Workup During Hospitalization.

Tests	Value	Reference range
Tryptase	<1	<11
Tryptase	1.2	<8.4
C1q	4.5	
C3 complement	138	88-193 mg/dL
C4 complement	24	15-57 mg/dL
IgE	<46	≤114 KU/L
Eosinophil count	0.5	
RAST for beef, chicken, lamb, and pork	Negative	
Aspiration bone marrow biopsy	Negative	
Flow cytometry	No immunophenotypic abnormalities	
β-1 globulin	0.5	0.4-0.6 g/dL
β-2 globulin	0.3	0.2-0.5g/dL
Anticardiolipin IgA	<11	<11
ANA	1:160, diffuse pattern	
ANCA	Negative	
Vitamin B ₆	28.5	20-125 pg/mL
Vitamin B ₁₂	575	232-1245 pg/mL
HIV	Negative	
Hepatitis C antibody	Nonreactive	
Hepatitis B surface antigen	Nonreactive	
Hepatitis B surface antibody	Reactive	
Urine drug screen	Negative	
Thyroid-stimulating immunoglobulin	89	<140% baseline
Salicylate	<3	<30 mg/dL
5-HIAA	2.6	≤114 kU/L
Anti-Rnp	Negative	
U1 Rnp Igg	0.6	≤4.9 U/mL
Sm Igg	3.4	≤6.9 U/mL
Anti-Sm	Negative	
Lyme (IgG and IgM)	Negative	
Anti-SSA (Ro)	<0.3	≤6.9 U/mL
Anti-SSB (La)	<0.3	≤6.9 U/mL

Abbreviations: IgE, immunoglobulin E; RAST, radioallergosorbent test; ANA, antinuclear antibody; ANCA, anti-neutrophil cytoplasmic antibody; HIV, human immunodeficiency virus; 5-HIAA, 5-hydroxyindoleacetic acid; Rnp, ribonucleoprotein; Sm Igg, Smith immunoglobulin; anti-SSA, anti-Sjogren-syndrome-related antigen A autoantibodies; anti-SSB, anti-Sjogren-syndrome-related antigen B autoantibodies.

the average cost for evaluation of patients with anaphylaxis to be \$30 545 per patient.⁷ Another case of an adolescent girl with multiple admissions for anaphylaxis reported a total hospital cost of \$243 000.⁵ Our patient underwent multiple workups at different times, which was tedious and time consuming, thus increasing the health care costs. Challenge tests with placebo followed by allergens should be done in suspected cases. Disclosure of negative results may often end up with confrontation and should be handled appropriately for better outcome.^{2,4} Early consultation with a psychiatry team and starting psychological counseling after making the diagnosis would prevent future hospital visits.^{2,6} Due to these patients' noncompliance and resistance to treatment, even psychiatric referral and psychotherapy have not been conclusively curative, which is one of the major challenges.^{1,2,5,7}

Conclusion

Factitious anaphylaxis can present as a life-threatening condition, mimicking a true anaphylaxis that needs immediate medical attention. A better approach to these patients would be a thorough clinical evaluation and early multidisciplinary involvement including internist, allergic specialist, and psychiatrist to avoid inappropriate intervention and cost. Long-term management goals should emphasize supportive care and regular follow-up to ensure the patient's mental health and overall well-being. So far, to our knowledge, there is very limited evidence of therapeutic intervention in managing FD. This case study highlights the importance of further evidence-based studies in the field of factitious disorder to decrease the disease burden and reduce the health care cost.

Declaration of Conflicting Interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.

Ethics Approval

Our institution does not require ethical approval for reporting individual cases or case series.

Informed Consent

Verbal informed consent was obtained from the patient(s) for their anonymized information to be published in this article.

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