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Case report

Anaphylaxis caused by mosquito allergy in systemic mastocytosis: A causal relationship

Anaphylaxie provoquée par une piqûre de moustique au cours d'une mastocytose systémique : une relation causale

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Abstract

Anaphylaxis to the mosquito is rare. We report 4 cases of patients with severe anaphylactic reaction after mosquito bites. These 4 patients have systemic mastocytosis. This combination of 2 rare diseases does not seem to be the result of chance and we recommend in case of anaphylaxis to the mosquito to carry out an assay of serum tryptase intercritical and in case of elevation to address the patient to our colleagues internists to search for a mastocytosis.

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Keyword: Mosquito allergy; Anaphylaxis; Systemic mastocytosis

Résumé

L'anaphylaxie aux piqûres de moustiques est rare. Nous rapportons 4 observations de réactions anaphylactiques sévères survenues après piqûre de moustique. Ces quatre patients étaient porteurs d'une mastocytose systémique indolente. L'association de ces deux pathologies exceptionnelles ne semble pas être le fait du hasard et nous recommandons en cas d'anaphylaxie aux piqûres de moustique de doser systématiquement la tryptase sérique à l'état basal et en cas de résultat élevé d'adresser le patient vers un centre de référence de la mastocytose.

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Mots clés : Allergie au moustique ; Anaphylaxie ; Mastocytose systémique

1. Abbreviations

SM systemic macrocytosis
PT prick test
sIgEs specific IgE serum

2. Introduction

Adverse reactions to mosquito bites have been recognized for some time. These usually consist of large local swelling and redness, generalised urticaria and angioedema. However, anaphylactic reaction is extremely rare [1,2]. We reported 4 patients who experienced systemic anaphylaxis from mosquito bites.

3. Case 1

In 2007, a 20-year-old man presented flush on his face associated with nausea, vomiting and dizziness after a mosquito

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Table 1
Characteristics of 4 patients.

	Case 1	Case 2	Case 3	Case 4
Prick test ^a	5/25	5/20	0/0	2/15
Specific IgE to mosquito (kUA/L) ^b	0.66	2.53	<0.10	0.18
Basophil activation response (%) ^a	NA	76	71.5	88.8
Tryptase (μg/L) on daily basis ^c	89	23.8	51.3	20.3

NA: not applicable.

^a Allergenic extract of *Aedes Communis* – (Stallergènes, France).

^b ImmunoCAP *Aedes Communis* (ThermoFisher, Uppsala, Sweden).

^c Normal range < 13.5 μg/L.

bite. The same year, he presented a couple of mosquito bites on his face. About 5 minutes after, he had the same reaction with hypotension. He was improved 30 minutes after he took an antihistaminic medication. The patient's skin prick test (PT) and specific IgE serum to mosquito (sIgE) were positive (Table 1).

4. Case 2

In the summer of 1996, a 32-year-old man presented a dozen of mosquito bites, while he was fishing. About 10 minutes later, he had generalized urticaria, facial angioedema associated with diarrhea, nausea, difficulty breathing and hypotension without lost consciousness. He was hospitalized and felt better quickly after corticosteroid IV injection. He has been bitten in the past and he had presented only localized swelling. He was bitten again one year later (unidentified insect). About 5 minutes later, he had a diffuse itchiness, sweating, difficulty breathing. He was improved by an injection of corticosteroid and adrenalin. Because of delayed resuscitation, he had hypoxic brain damage resulting in pyramidal syndrome. His allergy assessment confirmed sensitization to mosquito bites (Table 1).

5. Case 3

A 47-year-old man presented pruritus on his ankles after a non-identified insect bites, following by, 2 hours after, a loss of consciousness. Despite he had negative PT and specific sIgE, he showed a very strong basophil activation response to mosquito (Table 1).

6. Case 4

A 56-year-old woman felt dizzy and nauseous after mosquito bites since she was a child. Recently, in 2014, she presented an anaphylactic reaction (loss of consciousness, and urticaria) after multiple mosquito bites. Her allergy assessment was also positive (Table 1).

All of them were diagnosed with an indolent systemic mastocytosis (SM), according to the WHO diagnostic criteria for SM. SM was suspected on a high tryptase level, without any anaphylactic reaction (Table 1). The diagnosis was confirmed by abnormal proliferation of mast cells with abnormal expression of CD25 in bone marrow examination and a positive

activating mutation of KIT, D816 V. They did not benefice of any specific treatment, except an emergency kit with adrenaline. Immunotherapy with mosquito extract (*Aedes Communis* – allergenic extract by Stallergènes, France) was proposed for all of them. Three out four had a good response with afterward, mosquito bites without any systemic reaction. For the case 4, immunotherapy was stopped because of systemic reaction after each injection.

We reported a case series of 4 cases of anaphylaxis to mosquito bite and SM. In literature, there is just only one case report on this association, published in 2013 in the Lancet [3]. Reiter et al. described the case of a 56-year-old man who presented anaphylaxis after mosquito bite. As our second case, he had neurological defect. His bone marrow biopsy confirmed the diagnosis of indolent SM but he had previously maculopapular cutaneous mastocytosis. In our patients, the diagnosis of SM was evoked only on anaphylaxis caused by mosquito allergy, because none of our patient had cutaneous signs of SM.

Using immunotherapy with mosquito extract is still debated. Some authors considered that is not safe because of the unpredictable reaction [3]. In France, *Aedes Communis* is the only alternative for skin prick test, basophil release and immunotherapy with whole body extracts. As it was described previously in others studies [4,5], immunotherapy in our patients gives good results of protection that induces good tolerance and has very good efficacy. However, because of the end of production and commercialization of allergenic extract, we cannot do skin tests or desensitized patient anymore.

To conclude, SM and anaphylactic reaction to mosquito bites is not a fortuitous association. We wanted to emphasize the particular clinical and biological presentation such as atypical clinical reaction (rare cutaneous symptoms, frequent loss of consciousness) and weakness of the PT and sIgE. These characteristics are similar to what we know in allergy to Hymenoptera venom associated with mastocytosis.

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Disclosure of interest

The authors declare that they have no competing interest.

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