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Simultaneous Oral Mite Anaphylaxis (Pancake syndrome) in a Father and Daughter and a

**Review of the Literature** 

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polvo de casa.

Oral mite anaphylaxis (OMA), also known as pancake syndrome, is a condition characterized by

(severe) allergic reactions after ingestion of food containing mite-contaminated flour. We report for

the first time a case of OMA occurring simultaneously in a father and daughter that was probably

caused by mite-contaminated beignet flour.

An 18-year-old girl (patient 1) and her 48-year-old father (patient 2) attended our outpatient clinic

because of possible hypersensitivity reactions within 15-30 minutes after ingestion of homemade

apple beignets and coffee. Patient 1 experienced dyspnea, generalized pruritus, and eyelid

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angioedema. Patient 2 presented similar—albeit more severe—symptoms and had generalized urticaria. Both were successfully treated with antihistamines and short-acting  $\beta_2$ -agonists. The clinical history revealed that both patients had an atopic background with rhinoconjunctivitis and mild asthma due to monosensitization to house dust mite. On 1 occasion, patient 2 experienced an identical reaction after ingestion of pancakes made from the same packet of beignet flour, which was stored open at room temperature in a kitchen cupboard. Interestingly, the mother and another daughter, neither of whom was sensitized to house dust mite, had eaten the apple beignets without incident. The table summarizes the laboratory and skin test findings that demonstrate sensitization to house dust mite and various storage mites in both patients. In contrast, no sensitization was observed with wheat, buckwheat, lupine,  $\alpha$ -amylase, uncontaminated beignet flour, and yeast. Although the suspected beignet flour was thrown away and therefore unavailable for further testing, we believe these 2 simultaneous case histories and the absence of symptoms in 2 relatives are highly indicative of OMA caused by mite-contaminated beignet flour. Moreover, since the diagnosis of OMA was confirmed, both patients have eaten beignets made from uncontaminated beignet flour such as that applied in the skin tests.

Although the aeroallergens house dust mite and storage mite are well-recognized causes of respiratory allergies such as rhinoconjunctivitis and asthma, their potential as food allergens remains less clear. OMA is a relatively new syndrome, which was first described by Erben et al [1] in 1993. It affects patients of all ages and both sexes and manifests with symptoms that vary depending on the site and extent of mast cell/basophil degranulation, which generally occurs within 15-60 minutes of ingestion. Although the clinical course may be self-limiting, most reactions are severe, with angioedema (also of the oropharynx with stridor) and involvement of the upper and lower respiratory tracts (e.g. rhinorrhea, nasal itching and/or congestion, dyspnea, wheezing, and chest tightness). Gastrointestinal symptoms frequently complete the clinical picture. Cardiovascular reactions and death are not excluded [2-6]. To date, OMA has not been observed to present as isolated oral allergy syndrome. An association has been proposed between OMA and hypersensitivity

to nonsteroidal anti-inflammatory drugs [2,7] or exercise; however, this association was absent in the cases we report and elsewhere [6]. The latter has been designated *dust mite ingestion—associated exercise-induced anaphylaxis* [8]. OMA has also been described after inhaling cooking vapors from a commercial pancake mix contaminated with the house dust mite *Dermatophagoides farinae* [9].

Although most cases have been reported in tropical and subtropical countries, where climatological conditions are favorable for mite growth (high temperature and relative humidity), the findings presented here and in other case reports [9,10] indicate that OMA can also occur in countries with a temperate climate. In these cases, mite infestation should be sought in inappropriate storage conditions at ambient temperature, as we report here. In fact, packets of opened flour should be stored in sealed containers in the refrigerator or freezer, where conditions are hostile to mite infestation.

The foods predominantly involved in OMA are pancakes (most common), beignets, sponge cakes, pizza, pasta, wheat bread, white sauce, and meat or fish dusted with wheat flour. In Japan, the food most commonly involved in OMA is okonomiyaki mix, whose ingredients contain wheat flour. Takoyaki mix, which is similar to okonomiyaki mix, is the second most prevalent cause of OMA in Japan [6]. The mite species involved are *Dermatophagoides pteronyssinus*, *D. farinae*, *Blomia tropicalis*, and the storage mites *Lepidoglyphus destructor*, *Tyrophagus putrescientiae*, *Thyreophagus entomophagus*, *Blomia freemani*, *Suidasia medanensis*, and *Aleuroglyphus ovatus*. All of these species can live indoors if the conditions are favorable. To date, the allergen(s) responsible for OMA remains elusive. As cooked and baked foods are able to trigger symptoms, it has been suggested that heat-stable components such as Der p 2 are involved in the pathogenesis of OMA [2]. The patients reported here are sensitized to both Der p 1 and Der p 2, but not to house dust mite tropomyosin (Der p 10).

The criteria for the diagnosis of OMA are the following: compatible symptoms occurring after ingestion of food prepared with contaminated flour; previous history of rhinitis, asthma, atopic

dermatitis, and/or food allergy; demonstration of IgE-mediated sensitization to mite allergens in vivo or in vitro; positive skin test result with the suspect flour; negative skin test result to wheat and to uncontaminated flour; clinical tolerance to food made with uncontaminated wheat flour; and identification of mites or mite allergens in the suspect flour. However, as reported here and elsewhere [6], the culprit food/flour might not always be available for skin testing or microscopic evaluation. The main differential diagnoses are with genuine wheat allergy and wheat-dependent exercise-induced anaphylaxis.

No cure has been found for OMA. Patients who are sensitized to mites should avoid ingestion of mite-infested food. To our knowledge, there are no data on the effects of mite immunotherapy on OMA.

In conclusion, OMA is easily overlooked and may be responsible for anaphylaxis where no obvious cause is identifiable (idiopathic anaphylaxis). Therefore, mites should be considered in patients presenting with mite sensitization and food-induced allergic reactions with no apparent allergy to the ingredients.

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Parameter	Patient 1	Patient 2	Normal Value
	Total and Specific Ig	<u>l</u> E	
Total IgE	361.7	235.6	<120 kU/L
Dermatophagoides pteronyssinus	76	24.00	<0.35 kU <sub>A</sub> /L
Der p 1	49.90	6.62	<0.35 kU <sub>A</sub> /L
Der p 2	41.20	11.80	<0.35 kU <sub>A</sub> /L
Der p 10	< 0.35	< 0.35	<0.35 kU <sub>A</sub> /L
Dermatophagoides farinae	56.40	12.80	<0.35 kU <sub>A</sub> /L
Acarus siroª	3.57	0.52	<0.35 kU <sub>A</sub> /L
Glycyphagus domesticus <sup>a</sup>	3.40	1.28	<0.35 kU <sub>A</sub> /L
Tyrophagus putrescentiae <sup>a</sup>	12.00	1.56	<0.35 kU <sub>A</sub> /L
Wheat	<0.35	<0.35	<0.35 kU <sub>A</sub> /L
Buckwheat	<0.35	<0.35	<0.35 kU <sub>A</sub> /L
α-Amylase	<0.35	<0.35	<0.35 kU <sub>A</sub> /L
Lupine	<0.35	<0.35	<0.35 kU <sub>A</sub> /L
Saccharomyces cerevisiae	<0.35	<0.35	<0.35 kU <sub>A</sub> /L
	Skin Prick Tests <sup>b</sup>		
House dust mite	7/22	5/12	<3/3 mm
Wheat	0/0	0/0	<3/3 mm
Beignet flour (new packet, uncontaminated)	0/0	0/0	<3/3 mm