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Lobonemoides robustus Stiasny (jellyfish) anaphylaxis without poly-γ-glutamic acid sensitization

We read with great interest the two articles entitled “Anaphylactic shock after the ingestion of jellyfish without a history of jellyfish contact or sting” [1] and “Anaphylaxis caused by ingestion of jellyfish” [2] published in this journal. The former case of anaphylaxis occurred after *Cephea cephea* ingestion without any history of jellyfish sting [1], and the latter occurred after *Rhopilema esculentum* ingestion, following sensitization through the skin by jellyfish stings [2]. Oral jellyfish challenge tests or skin-prick tests for poly-γ-glutamic acid (PGA) and natto (fermented soybean) were not conducted in either study. Here, we describe a case of anaphylaxis following *Lobonemoides robustus* (LR) ingestion, without any history of jellyfish sting, in which an oral jellyfish challenge test and skin-prick tests for jellyfish, PGA, and natto were administered. A 14-year-old Japanese boy developed wheezing and dyspnoea 1 hour after he had dinner at home, which included salted jellyfish (LR), and was rushed to our hospital. He had no history of jellyfish contact or stings. On arrival, he was alert, with a blood pressure of 124/74 mm Hg, oxygen saturation level <85%, bilateral bloodshot eyes, nasal discharge, and erythema. He was diagnosed with anaphylaxis. His symptoms resolved after treatment

with intramuscular adrenaline, inhaled procaterol, and intravenous hydrocortisone. Four weeks later, we administered an oral food challenge test to the patient for the same jellyfish. Forty-five minutes after having ingested the jellyfish (30 minutes after initially ingesting 2 g plus 15 minutes after additional ingestion of 6 g), he experienced the same symptoms. We diagnosed the patient with anaphylaxis following jellyfish ingestion. His symptoms resolved with the same treatment, as before. Skin-prick testing for PGA (20 mg/mL) and prick-to-prick tests with the same jellyfish and natto were also performed. The jellyfish yielded a positive result at 15 minutes with a wheal diameter of 5×5 mm, whereas the other results were negative (positive control: 10 mg/mL histamine hydrochloride, with a wheal diameter 10×10 mm; negative control: saline, yielding no wheal). The patient was diagnosed with anaphylaxis due to jellyfish ingestion, but not PGA. There are eight case reports of anaphylaxis following jellyfish ingestion, including ours, in the literature [1-7]. Seven were reported by Japanese physicians [1-6] and five were published after 2017 [3, 5-7]. Although anaphylaxis following jellyfish ingestion has been thought to be rare, reports have rapidly increased recently, especially in Japan, probably due to increased knowledge. Anaphylaxis of unknown cause might include jellyfish anaphylaxis. Regarding the actual type of jellyfish ingested, LR (our case), *Cephea cephea* [1], *Rhopilema esculentum* [2, 6], *Stomolophus meleagris* [3], and *Mastigias papua* [5] have been reported (figure 1). Positive skin-prick test results with LR (our case), *Cephea cephea* [1], *Rhopilema esculentum* [2, 5], *Stomolophus meleagris* [3], *Mastigias papua* [5], and *Nemopilema nomurai* [5] have also been reported (figure 1). In contrast, *Stomolophus meleagris* and *Lobonema smithi* were reported to show negative results on skin-prick testing [5] (figure 1). The results of skin-prick tests for several kinds of jellyfish suggest that the causative antigen may differ for each jellyfish. The oral jellyfish challenge tests with LR (our case) and *Mastigias papua* [5] have been reported, both yielding positive results (figure 1). Five cases have been reported with a history of contact with or stings by jellyfish [2-4, 6, 7].

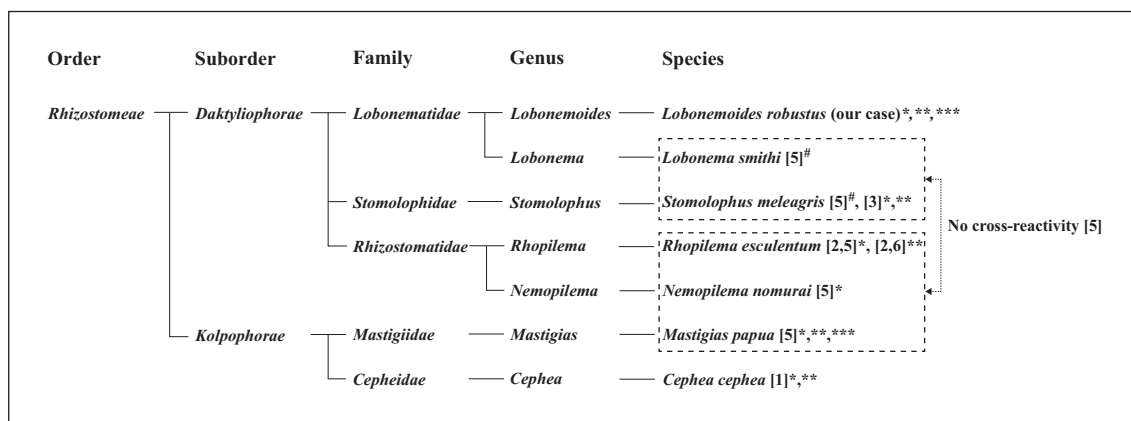


Figure 1. Taxonomic classification of jellyfish associated with anaphylaxis following ingestion. There has been no report of anaphylaxis following ingestion of *Lobonema smithi* or *Nemopilema nomurai*.
[#]Negative skin prick test result. *Positive skin prick test result. **Actual type of jellyfish ingested. ***Positive oral challenge test.

The mechanism of jellyfish allergy remains unknown. In a report of a subject with allergy to jellyfish and natto [4], the authors proposed the possibility of epicutaneous sensitization to PGA, a major allergen of natto, due to jellyfish stings. However, a skin-prick test for PGA, a jellyfish sting component, yielded a negative result. No cross-reactivity between jellyfish and natto was demonstrated in our patient, suggesting that the patient was sensitized to allergens, other than PGA, via different routes derived from jellyfish stings. To the best of our knowledge, this is the first reported case of anaphylaxis following LR ingestion, for which both the oral challenge test and skin-prick test were administered. Our results suggest that not only *Cephea cephea*, *Rhopilema esculentum*, *Stomolophus meleagris*, or *Mastigias papua*, but also LR ingestion can lead to anaphylaxis. Our patient with jellyfish anaphylaxis was sensitized to allergens, other than PGA, via different routes derived from a jellyfish sting or natto.

Informed consent for publication was obtained from the patient and his parents. ■

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Xanthoma disseminatum with rare extensive lesions

A 38-year-old man presented with a 10-year history of multiple cobblestone-like, egg-sized lesions involving the

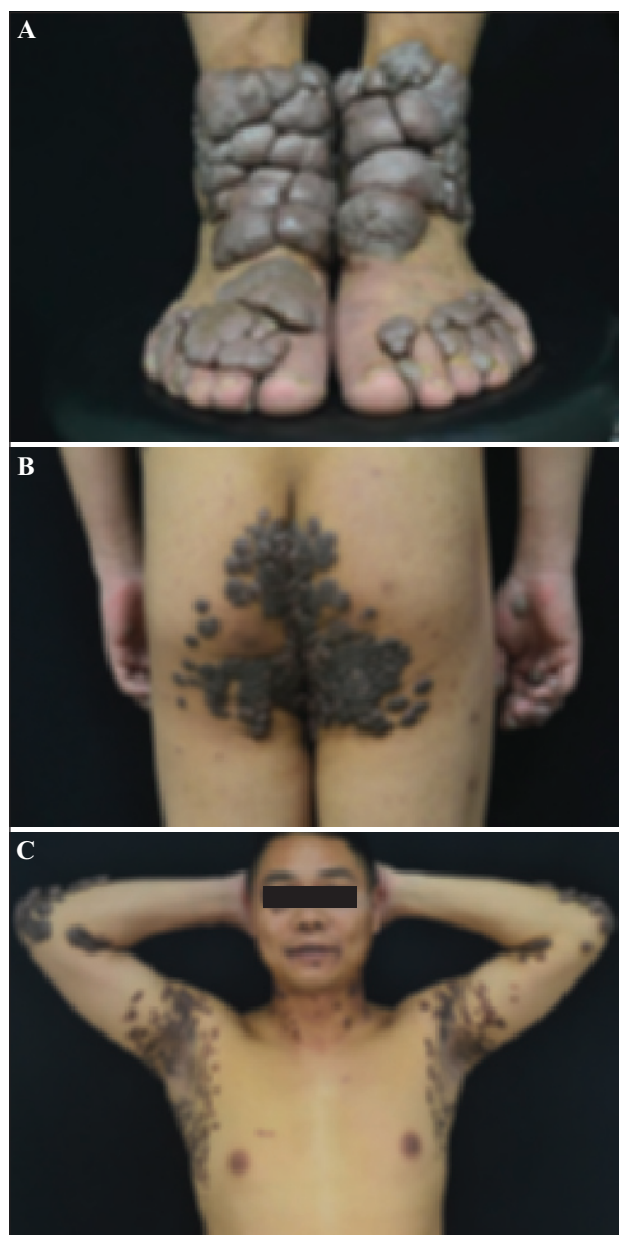


Figure 1. Clinical images of cutaneous eruptions concentrated at flexural sites; papules and plaques are present on ankle and interdigital regions (A), the buttocks (B), axillae (C), and neck (D). E) Hematoxylin and eosin staining ($\times 200$) showing dermal infiltration of numerous irregular macrophages with foamy cytoplasm, associated with lymphocytes and multinucleated giant cells (arrow). F) Xanthoma disseminatum cells stain ($\times 200$) strongly positive for the CD68 marker.

face, flexures of the trunk, and extremities (figure 1), with no systemic symptoms. The lesions first appeared as yellowish, well-demarcated papules, especially in the flexures of the trunk, which then increased progressively in size and number. Lesions were also observed on the nasal and labial mucosa. The patient had previously been treated with CO₂ laser in 2011 and surgical resection in 2013 to remove his facial lesions, but these relapsed after one year. The lesions on the trunk and extremities worsened progressively into their current state without any interven-