

Recurrent Erythema after Eating in a 1-year-old Boy: A Quiz

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A 1-year-old boy presented to the dermatology clinic for recurrent erythema on his right cheek after eating solid food. It was first observed 6 months earlier when he was given pureed apple. It could easily be triggered with a variety of acidic and sweet fruits and foods that need more mastication. The erythema appeared within a few seconds after eating and resolved spontaneously in about 30 min, with the same distribution on each occurrence (Fig. 1). There was no pruritus, pain, sweating, or associated systemic symptoms such as vomiting and diarrhoea. The boy was born at full term and had a history of right facial forceps injury at birth

(Fig. 2). He had no prior history of urticaria or eczema, and was in good health.

What is your diagnosis?

Differential diagnosis 1: Frey syndrome

Differential diagnosis 2: Food allergy

Differential diagnosis 3: Mastocytosis

Differential diagnosis 4: Port wine stains

See next page for answer.



Fig. 1. Erythema appeared on the right cheek after eating.



Fig. 2. Patient had a history of right facial forceps injury at birth.

ANSWERS TO QUIZ

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Diagnosis: Frey syndrome

Frey syndrome (FS), or gustatory sweating, usually occurs in adults who undergo salivary gland, neck, or facelift surgery, or those suffering from diabetes mellitus. However, it is rare in the paediatric population. The case we reported



Fig. 3. Hyperpigmentation seen at 1-year follow-up.

had a history of forceps injury on the right cheek at birth, which may be the most common cause of FS in infancy. A national retrospective study found that 68% of patients with infantile FS had visible marks at birth following a traumatic instrumental delivery (1). Another systematic review indicated that 52% of 121 individual cases of FS, not associated with surgery or diabetes, had histories of forceps birth (2).

The most widely accepted mechanism for development of FS is that it results from aberrant reinnervation of cholinergic sympathetic sweat glands and blood vessels in the skin with parasympathetic fibres from the auriculotemporal nerve that have been exposed or injured (3). Unlike FS in adults, sweating is uncommon in children. The exact cause is not well understood but may be related to the less intense regeneration after forceps injury, rather than nerve section.

It was reported that 20% of infantile FS cases were completely relieved spontaneously within 11 to 81 months after the appearance of symptoms (1). In addition, 57% of cases showed regression during the same period (1). Treatment is considered ineffective and unnecessary. In our case, the symptom persisted at the 1-year follow-up, with hyperpigmentation on the same area of the right cheek (Fig. 3).

Infantile FS is often misdiagnosed as food allergy because it usually occurs when infants start consuming a wider variety of foods. The characteristics of infantile FS include being triggered by various foods, involving the same skin area on each occurrence, the absence of pruritus, and without multisystem involvement. Thus it can be distinguished from food allergy. Dermatologists should be familiar with infantile FS to avoid unnecessary diagnostic procedures and restricted diets.

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