

## CASES FOR DIAGNOSIS

# Linear Erythema on the Face of a Boy

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## Clinical History

The patient was a 17-month-old boy who was seen for an episodic, transient facial rash (lasting approximately 10 minutes) that had been occurring since 4 months of age and was related to eating certain fruits and, later, other foods.

Pregnancy and delivery were normal, with no complications, and the infant had received maternal milk exclusively until 4 months of age, when his mother started to introduce other foods.

## Physical Examination

On physical examination, a linear erythema developed between the commissure of the mouth and the ear on the left side 10 seconds after eating one of the causative foods; spontaneous improvement occurred after 10 minutes (Figure 1).



Figure 1.

**What Was the Diagnosis?**

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## Diagnosis

Frey or auriculotemporal syndrome.

## Clinical Course and Treatment

The benign, self-resolving nature of this syndrome was explained to the parents.

## Comments

Frey or auriculotemporal syndrome, first described by Duphenix in 1757 and published by Frey in 1923,<sup>1</sup> is characterized by recurrent episodes of erythema, sweating, or both, in the distribution of the auriculotemporal nerve, usually unilaterally, in response to gustatory and, occasionally, tactile stimuli.<sup>2-5</sup>

In adults it is a common sequela of surgery, nonsurgical trauma, or infection of the parotid gland.<sup>2</sup> It is a rare condition in children, with less than 40 cases reported in the literature,<sup>4</sup> and, in the majority of cases, onset occurs after the introduction of solid foods into the diet. In a case series of children, a history of delivery assisted by the use of forceps applied to the head was found in more than 50% of cases, suggesting that trauma to the area of the parotid gland could be a causative factor in affected children.<sup>2,6</sup>

The auriculotemporal nerve, which runs towards the temporal region, carries sensory fibers from the preauricular and temporal regions, as well as sympathetic and parasympathetic fibers. The secretory fibers of the parasympathetic branches run to the parotid gland, while the sympathetic fibers innervate the subcutaneous arterioles and the eccrine glands. The probable mechanism of auriculotemporal syndrome is an incorrect regrowth of parasympathetic fibers along the sympathetic pathways during nerve regeneration after trauma. Regeneration of the fibers along an incorrect path towards the sweat glands causes sweating, whereas regeneration towards the cutaneous blood vessels produces food-related erythema. The use of forceps in more than 50% of cases reported in children suggests that trauma during delivery may damage this nerve, leading to the formation of an aberrant nerve pathway during the regeneration process. The onset of symptoms occurs several months after trauma (usually 3-6 months), which is probably due to the time required for the nerve to regenerate.<sup>2,5</sup>

In some cases there is no known history of trauma in the region of the parotid gland; an alternative explanation is the presence of a congenital aberrant cranial nerve.<sup>3</sup> It is also possible that very vigorous mastication, particularly with favorite foods, could lead to more intense stimulation of the parotid gland than sucking during breast-feeding.<sup>2</sup>

The diagnosis is clinical and is confirmed by an oral provocation test. The main differential diagnosis is with food allergies; in contrast to allergy, the administration of antihistamines before eating or the exclusion of certain foods does not improve the symptoms. In addition, the syndrome is not associated with pruritus, gastrointestinal symptoms, urticarial lesions, or angioedema, and it is usually unilateral.<sup>6</sup>

With regard to treatment, the administration of anticholinergic agents or botulinum toxin has improved the gustatory sweating in some cases; however, these treatments are of no help in cases of facial erythema, the sign that predominates in infants. Moreover, botulinum toxin is not metabolized in infants and can cause serious toxic effects; its use is therefore not recommended in this age group. As it is a benign condition in children, and usually resolves gradually, no treatment is required.<sup>2,5,6</sup>

In conclusion, we present a case of Frey syndrome, a very rare condition in children, with a benign, self-limiting course. It must be recognized in order to avoid unnecessary tests and inappropriate treatments.

## Conflicts of Interest

The authors declare no conflicts of interest.

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