

Auriculotemporal syndrome following forceps delivery

Sarah Edgerley MD, Kara Robertson MD

■ Cite as: *CMAJ* 2023 January 9;195:E15. doi: 10.1503/cmaj.221178

An 8-month-old boy, born at term via vaginal delivery with forceps assistance, presented to our allergy clinic with symptoms concerning for fruit allergy. He was otherwise healthy, on no medications and without surgical history.

After the introduction of solid foods at 4 months, the patient developed intermittent flushing on the left side of his face with eating (Figure 1). He had no associated pain, urticaria, angioedema or systemic involvement. Facial flushing was invariably left-sided and it appeared only while eating. Symptoms occurred most predictably with fruit ingestion, but not with every exposure. The family removed presumed triggering foods from his diet but symptoms persisted. We diagnosed auriculotemporal (Frey) syndrome. We suggested foods be reintroduced and we deprescribed his epinephrine autoinjector.

Auriculotemporal syndrome is characterized by gustatory flushing, warmth and sweating of the facial area innervated by the auriculotemporal nerve.^{1,2} Damage to the nerve causes aberrant regeneration of parasympathetic fibres along sympathetic pathways, resulting in stimulation of cutaneous vasculature and sweat glands, instead of the salivary glands, after

gustatory stimuli.^{1,3} The differential diagnosis for facial flushing includes rosacea, food allergy, medication adverse effect and neuroendocrine causes.

In adults, auriculotemporal syndrome is usually secondary to facial trauma or otolaryngological surgery.^{1,2} In children, injury from forceps (as in this patient) or traumatic delivery commonly causes unilateral symptoms; in 1 study, 69% of cases resolved after infancy.¹ Symptoms are typically triggered by eating acidic, sour or spicy foods, which stimulate salivation.¹ Fruits are the most common initial triggers in children.¹ As initial presentation corresponds to age of food diversification, auriculotemporal syndrome is often misdiagnosed as food allergy, leading to unwarranted investigations and inappropriate elimination diets.¹⁻³ Auriculotemporal syndrome can be distinguished by the characteristic distribution of flushing, the absence of other immunoglobulin E-mediated symptoms, association with typical unrelated food triggers that are uncommon allergens, rapid symptom resolution without treatment and history of forceps delivery.^{1,3} Clinical history alone is sufficient to make the diagnosis of auriculotemporal syndrome, but can be aided by provocation tests.¹



Figure 1: An 8-month-old boy with unilateral, left-sided facial flushing that developed within a few seconds of eating.

References

1. Blanc S, Bourrier T, Boralevi F, et al.; Frey Syndrome Collaborators. Frey syndrome. *J Pediatr* 2016;174:211-7.e2.
2. Buyuktiryaki B, Sekerel BE. Is it food allergy or Frey syndrome? *J Allergy Clin Immunol Pract* 2015;3:269-70.
3. Els I, Delanty J. Infantile Frey syndrome. *J Paediatr Child Health* 2021; 57:2040-1.

Competing interests: None declared.

This article has been peer reviewed.

The authors have obtained patient consent.

Affiliation: Division of Clinical Immunology and Allergy, Department of Medicine, Western University, London, Ont.

Content licence: This is an Open Access article distributed in accordance with the terms of the Creative Commons Attribution (CC BY-NC-ND 4.0) licence, which permits use, distribution and reproduction in any medium, provided that the original publication is properly cited, the use is noncommercial (i.e., research or educational use), and no modifications or adaptations are made. See: <https://creativecommons.org/licenses/by-nc-nd/4.0/>

Correspondence to: Kara Robertson, kararobertson@dal.ca