Auriculotemporal syndrome associated with a traumatic vaginal delivery at birth

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DESCRIPTION

Case

A middle childhood female patient presented to the paediatric dermatology clinic for evaluation of a recurrent and reproducible right-sided facial rash that started when she was an infant. The patient was delivered by traumatic vaginal delivery at term, with failed vacuum extraction progressing to a forceful forceps-assisted vaginal delivery. The patient was admitted to the neonatal intensive care unit after developing a cephalo-haematoma that led to prolonged jaundice and required phototherapy. The patient’s dentist also reported ipsilateral hypomineralisation of the molar teeth, requiring two crowns and five fillings by early childhood.

The patient’s mother was induced at 38 weeks’ gestation. The patient’s birth weight was 3.6 kg, length 53 cm and head circumference 35.5 cm. The fetal presentation before birth is not known. There was no history of broader musculoskeletal or orthopaedic issues such as leg bowing, abnormal skull shape or fractures. Her development is otherwise normal.

On mastication of food, the patient developed a well-demarcated erythematous eruption on the right temple, extending down to the malar eminence and oral commissure (figure 1). The rash was not associated with urticaria, pain, pruritus or hyperhidrosis. The rash self-resolved over a few minutes after finishing eating. The phenomenon would occur independent of the type of food being consumed and was readily reproducible. The patient’s clinical findings were consistent with auriculotemporal syndrome, also known as Frey’s syndrome.

Written informed consent for patient information and images to be published was provided by the subject(s) or a legally authorised representative.

DISCUSSION

Auriculotemporal syndrome describes the transient erythema and/or perspiration over the distribution of the auriculotemporal nerve, secondary to parasympathetic nerve activation.1 The use of forceps in the patient’s birth may have caused auriculotemporal nerve damage as well as contributed to enamel hypoplasia.2

The proposed mechanism of auriculotemporal syndrome is a collateral connection of parasympathetic fibres into vacated sympathetic pathways.3 Often a result of surgery or trauma, postganglionic parasympathetic fibres of the auriculotemporal nerve and sympathetic nerves innervating nearby subcutaneous sweat glands and blood vessels are damaged.3 Neurotrophic factors released by damaged sweat glands, such as neurturin, may play a role in attracting regenerating parasympathetic fibres. The end result is a connection between the auriculotemporal parasympathetic secretomotor and vasodilator fibres with the overlying sympathetically denervated eccrine sweat glands and blood vessels.3 During episodes of acetylcholine release from parasympathetic nerves, typically during salivation while eating, the eccrine sweat glands perspire and cutaneous blood vessels vasodilate, attributing to the symptoms of auriculotemporal syndrome.1

Auriculotemporal syndrome may be mistaken for a food allergy due to the onset of cutaneous symptoms coinciding with mastication. This can often lead to unnecessary elimination diets before the correct diagnosis is made.4 It is therefore incumbent on clinicians to delineate the two conditions.

Figure 1 A well-demarcated erythematous eruption on the right temple, extending down to the malar eminence and oral commissure.
Rashes related to food allergies typically involve the bilateral face and are accompanied by other symptoms such as urticaria, angioedema, respiratory and gastrointestinal symptoms. Auriculotemporal syndrome is almost exclusively unilateral. Patients also report a traumatic instrumental delivery or trauma to their area (e.g., surgery to the parotid gland, fractures to mandibular condyle or temporomandibular joint).

Treatment of auriculotemporal syndrome may be sought due to social concerns for profuse flushing and sweating when eating. Botulinum toxin may be used as a first-line treatment option. Botulinum toxin acts by transiently inhibiting the release of acetylcholine from nerve terminals, with therapeutic effects lasting up to 9–12 months.

**Learning points**

- A diagnosis of auriculotemporal syndrome in a paediatric patient is favoured over food allergies when the patient has a history of traumatic instrument delivery at birth or trauma to the parotid area, presents with a unilateral rash over the distribution of the auriculotemporal nerve and lacks systemic symptoms.
- Auriculotemporal (or Frey) syndrome describes the transient erythema and/or perspiration over the distribution of the auriculotemporal nerve on parasympathetic nerve activation, typically during mastication.
- Botulinum toxin inhibits acetylcholine release from nerve terminals, which may help with the symptoms associated with auriculotemporal syndrome such as flushing or perspiration.