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CASE REPORTS

Frey's Syndrome in Childhood

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Abstract. - Frey's syndrome, also known as auriculotemporal syndrome is characterized by recurrent episodes of facial flushing and/or sweating along the distribution of the auriculotemporal nerve, that occurs in response of gustatory stimuli. The disorder is rare in children. We report a 4-year-old girl with flushing without hyperhydrosis in the left cheek on eating.

Key words: Frey's syndrome, auriculotemporal syndrome, childhood.

SÍNDROME DE FREY EN LA INFANCIA

Resumen. El síndrome de Frey o auriculotemporal se caracteriza por la aparición de episodios recurrentes de eritema facial y/o hiperhidrosis, localizados en el territorio del nervio auriculotemporal, que ocurre en respuesta a los estímulos gustatorios. Este cuadro es poco frecuente en la infancia. Presentamos el caso clínico de una niña de 4 años de edad que presentaba eritema sin hiperhidrosis en la mejilla izquierda tras la masticación.

Palabras clave: síndrome de Frey, síndrome auriculotemporal, infancia.

Introduction

Frey syndrome is characterized by gustatory hyperhidrosis and localized flushing in the region of the auriculotemporal nerve. It is a common complication in adults following parotidectomy or trauma, but it is rare in children. We report a case of auriculotemporal syndrome in a 4-year old girl.

Case Description

The patient was a 4-year old girl, born full-term by vaginal delivery, with no other relevant personal or family history. She attended our clinic because she had presented with slight flushing on the left cheek a few seconds after mastication ever since she was 6-months old. This would resolve spontaneously and was not associated with other symptoms. According to her parents, this flushing appeared with many foodstuffs and especially with candy and fruit. A challenge test was carried out by giving the girl a piece of candy to suck, and a few seconds later a line of mild flushing was seen extending from the edge of the mouth to the left temporal region (figure). Within minutes, the intensity had decreased.

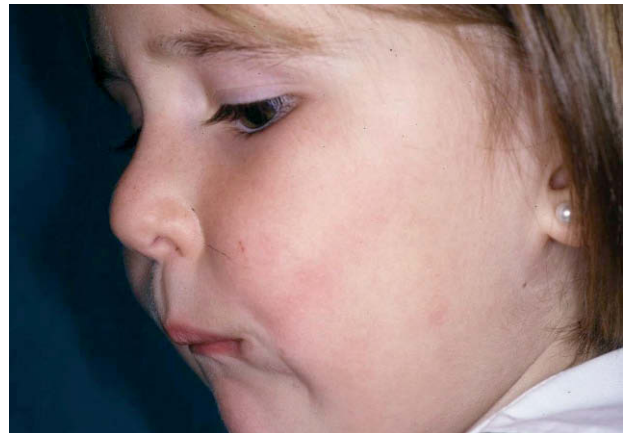

Figure. Mild patch of flushing in a line across the left cheek, appearing a few seconds after ingestion of a piece of candy.

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Discussion

Auriculotemporal syndrome was first described in 1757 by Duphenix and published by Frey in 1923.[1] It is characterized by the appearance of flushing, hyperhidrosis, or both, in the region of the auriculotemporal nerve, occurring in response to gustatory and occasionally tactile stimuli. Unlike adults, flushing is the most characteristic symptom in children, with few reports of sweating. The symptoms are first observed when solid food is introduced (in the first year of life). They begin a few seconds after food is ingested and resolve spontaneously within 30 to 60 minutes.[2] Frey syndrome is rare in childhood. On reviewing the literature, we found fewer than 40 case reports, 4 of which were bilateral.[3,4] The pathophysiology of this syndrome is unknown. The most accepted theory is that the syndrome is caused by irregular healing following trauma, which results in aberrant nerve regeneration, with parasympathetic fibers in contact with sympathetic fibers. In approximately half of the published cases the child was born by forceps delivery, and in patients with bilateral symptoms, only one of them was a forceps delivery. In the cases where forceps were not used and no trauma injury was apparent in the area of the parotid gland, the proposed mechanism is an anomalous congenital nervous connection.[5]

Many cases of Frey syndrome in childhood are wrongly diagnosed as food allergies. Treatment of adults has ranged from anticholinergic drugs or botulinum toxin to dorsal sympathectomy or resection of the auriculotemporal nerve; however, most treatment methods have proved ineffective and have sometimes produced significant morbidity. In children none of these treatments is recommended as this is a benign, nonprogressive condition that may resolve spontaneously.[3] Hence, correct diagnosis of this symptom is important in order to avoid unnecessary examinations and ineffective treatments.

Conflict of interests

The authors declare no conflict of interests

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