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Incomplete auriculotemporal nerve syndrome – mimicry of oral allergy syndrome

Pollen allergies are commonly symptomatic as seasonal rhinoconjunctivitis. The majority of patients with pollen allergies develop an oral allergy syndrome due to cross-reactive homologous allergens in plant foods. Symptoms may vary from minor local oropharyngeal sensations and swelling to life threatening angioedema and glottis edema. We present the case of a 6-year-old male with suspected oral allergy syndrome who was referred for allergological work up. However, an incomplete auriculotemporal nerve syndrome was diagnosed, mimicking oral allergy syndrome.

Key words: pollen food allergy syndrome (oral allergy syndrome), auriculotemporal nerve syndrome, facial erythema

Oral allergy syndrome (OAS) summarizes allergic reactions to food allergens with coexisting allergies to aeroallergens [1]. These allergies are based on cross-reactive homologous allergens present in related and unrelated plant species [2]. Commonly, patients suffer from seasonal rhinoconjunctivitis and oropharyngeal itching or burning. Typically, symptoms start a few minutes after intake of uncooked plant foods.

Case report

An 6-year-old male presented with a 2-year history of recurrent episodes of unilateral swelling and erythema on his right cheek shortly after intake of distinct fresh plant foods such as pears, apples, kiwis or carrots. Symptoms lasted only for a short period of time and disappeared spontaneously. At the same time he had developed seasonal rhinoconjunctivitis.

The past medical history was unremarkable except for parotidectomy and lymphadenectomy after recurrent right-sided parotitis two years earlier.

On first presentation, the patient was asymptomatic. The prick tests to inhalant allergen sources and plant foods revealed positive wheal reactions of 4-6 mm and over 6 mm diameter to pollen allergens including grass, rye, birch, hazel, alder and red beech and 2-3 mm wheal reactions to anise, hazelnut, and caraway. The suspected plant foods pear, apple, kiwi and carrot were negative in the prick to prick test.

Provocation testing was performed. Open provocation with a tender pear did not provoke symptoms. However, chewing of a firm carrot revealed erythema at the right cheek (*figure 1*). Mucosal symptoms were absent. The iodine starch test as indicator of gustatory sweating was negative. The differential diagnosis of unilateral facial erythema and swelling (i.e. Quincke's edema, hereditary angioedema, acute contact dermatitis, erysipelas, N. flammeus [3]) could be ruled out clinically. In the context of the patient's status post right-sided parotidectomy and provocation of symp-

toms by intense chewing of firm foods, the unilateral erythema without mucosal symptoms was diagnosed as incomplete auriculotemporal nerve syndrome.

Discussion

Incomplete auriculotemporal nerve syndrome is an infrequent disease which can be misdiagnosed as oral allergy syndrome. Understanding of OAS is based on recent insight into the molecular properties of allergens and cross reactivities of homologous pollen and food allergens [2, 4-8]. Based on botanical and molecular relationships, marker allergens could be defined [7]. Members of the PR 10 subfamily of pathogenesis related proteins, including



Figure 1. A) Swelling and erythema with undefined borders on the right cheek after chewing of raw carrot. B) Hypertrophic scar at the right retroauricular region after parotidectomy.

the major birch pollen allergen Bet v 1 and homologous allergens from carrot, apple and pear [1, 6-8] could theoretically have been the relevant cross-reactive allergens in the case presented. However, in contrast to positive skin prick testing to tree pollen, including birch, hazel, alder (relevant for the patient's seasonal rhinoconjunctivitis) all the suspected plant foods (pear, apple, kiwi and carrot) were negative and the patient did not develop mucosal symptoms upon oral provocation. Further differential diagnosis of unilateral facial erythema and swelling could be excluded.

The auriculotemporal syndrome (syn. Frey's syndrome) was first described in the middle of the 18th century by Duphenix [9] and later named after the neurologist Lucia Frey [10]. It represents a local hyperhidrosis in the inner-

vation area of the N. auriculotemporalis after gustatory stimuli. It follows injury or surgery in the regio parotideo-masseterica, neck dissection or luxation of the temporomandibular joint, parotitis or parotid abscess. It may also be a rare manifestation of diabetic neuropathy or post-herpetic neuralgia and may be a sign of Horner's syndrome, which includes ipsilateral anhidrosis, ptosis, miosis and enophthalmus.

The onset of Frey's syndrome occurs typically 1 month to 5 years post surgery or injury in the preauricular region and affects 37 to 100% of patients with such conditions [11]. In our case, the clinical manifestation was first observed two years after parotidectomy.

The pathophysiology of this syndrome is not precisely elucidated. It most frequently results from an injury to the auriculotemporal nerve. In addition, an irritation of the N. auriculotemporalis by scar formation had been suggested. Under normal conditions, the auriculotemporal nerve carries sensory fibers from the skin, parasympathetic fibers to the salivary glands and sympathetic fibers to the sweat glands of the preauricular region (figure 2A). According to the misdirection hypothesis of the pathogenesis of auriculotemporal nerve syndrome, parasympathetic nerve fibers regenerate aberrantly into the sweat glands as well as blood vessels of the skin of the preauricular region (figure 2B). In case of concomitant damage to the greater auricular nerve the infraauricular region (figure 2C) is also affected. The gustatory sweating in Frey's syndrome is usually mild and is not necessarily induced by gustatory stimuli. Incomplete clinical variants of Frey's syndrome, as demonstrated in our case, may present without sweating, but erythema, as a result of aberrant parasympathetic nerve fibers being predominantly redirected to cutaneous blood vessels.

Similar to our case, in the only previously reported case, a 21-year-old woman complained of erythema, sweating and heat in the right cheek after intake of several foods such as chocolate, fruits and nuts. Her history revealed a jaw fracture 2 years earlier [12].

Due to the mild symptoms only about 10% of patients require treatment to reduce gustatory sweating [11, 13]. The therapeutic options of the auriculotemporal nerve syndrome include topical application of aluminium chloride hexahydrate 20% solution, intracutaneous injection of botulinum toxin A or oral intake of sage extract [14]. In the case presented no treatment and no elimination diet (previously recommended under the suspicion of OAS) was required. ■

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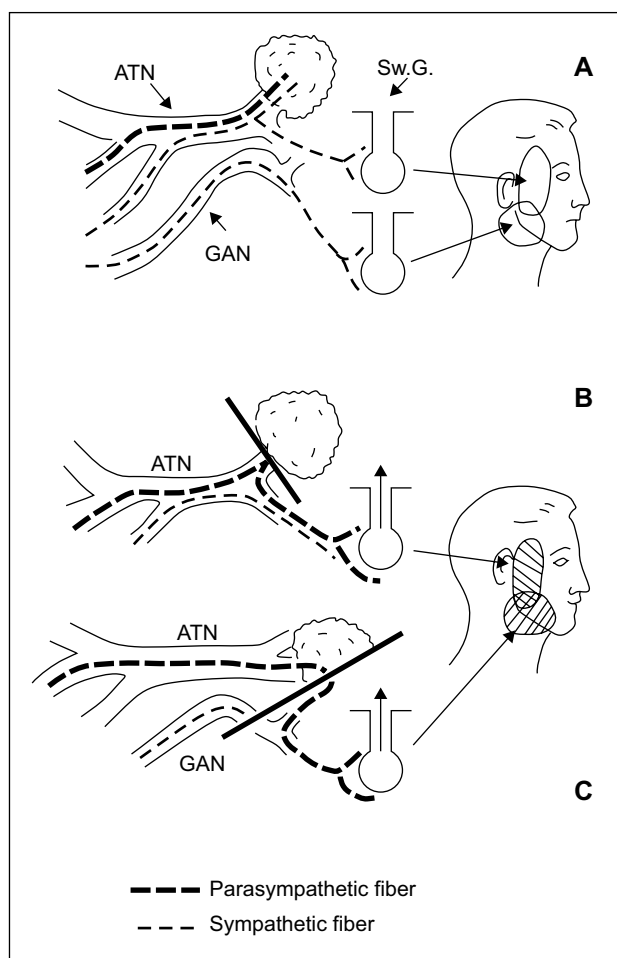


Figure 2. Scheme of parotid innervation modified from [11]: ATN auriculotemporal nerve, GAN greater auricular nerve, Sw.G. sweat gland. **A)** normal innervation. The auriculotemporal nerve carries sensory fibers from the skin, parasympathetic fibers to the salivary glands and sympathetic fibers to the sweat glands of the preauricular region. **B)** After surgery of the parotid gland or injury to the preauricular region, parasympathetic fibers in the auriculotemporal nerve regenerate and migrate into the postganglionic sympathetic fibers to reach the sweat glands as well as the blood vessels of the preauricular region. **C)** When the parotid gland and the greater auricular nerve are damaged, the regenerating parasympathetic fibers from the ATN migrate into the GAN to reach the sweat glands of the infraauricular region.

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