

Rare presentation of Lucie Frey syndrome after trigeminal herpes zoster

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Abstract

Lucie Frey Syndrome, or "gustatory sweating," is a rare autonomic neuropathy caused by aberrant parasympathetic reinnervation following nerve damage. While commonly associated with traumatic or surgical etiologies, infectious causes such as herpes zoster are exceptionally rare in pediatric cases. We report the case of a 3-year-old female who developed unilateral erythema and focal hyperhidrosis on the right hemiface.

Keywords: Lucie Frey Syndrome; Gustatory Sweating; Herpes Zoster; Pediatric Autonomic Neuropathy; Aberrant Nerve Regeneration

1. Introduction

Lucie Frey Syndrome, or "gustatory sweating," is an uncommon autonomic neuropathy resulting from aberrant parasympathetic reinnervation following nerve damage. While typically associated with traumatic or surgical etiologies in adults, infectious origins, such as herpes zoster, remain exceptional in pediatric cases.

2. Case presentation

We report the case of a 3-year-old female patient with a history of herpes zoster involving the right hemiface, treated successfully two years prior. She developed unilateral episodes of erythema and focal hyperhidrosis affecting the right cheek, triggered by food intake or chewing. These episodes, which resolved spontaneously within minutes, were objectively confirmed via a provocation test using salty foods. Comprehensive otolaryngologic evaluations, including rhinoscopy, computed tomography (CT) of the face, and soft tissue ultrasonography, revealed no abnormalities.

A diagnosis of Lucie Frey Syndrome was made based on the patient's clinical history, the characteristic topography of symptoms, and the positive provocation test (Figure 1).

This diagnosis underscores a rare infectious etiology in the pediatric age group. Symptomatic management was initiated with the application of topical anticholinergics, specifically oxybutynin cream, to mitigate hyperhidrosis. Additionally, botulinum toxin injections are planned to provide sustained control of symptoms.

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Figure 1 Clinical aspect of the patient after Provocation Test: Hyperhidrosis and Erythema of the Right Cheek

3. Discussion

Lucie Frey Syndrome, also referred to as "gustatory sweating," represents a unique challenge for dermatologists due to its rare presentation and underlying autonomic neuropathology. This condition, resulting from aberrant parasympathetic reinnervation, is primarily recognized in adults following surgical or traumatic injury to the auriculotemporal nerve. However, the pediatric occurrence of this syndrome, particularly with an infectious etiology such as herpes zoster, remains exceedingly rare and clinically significant (1).

From a dermatological perspective, the symptoms—episodic erythema and focal hyperhidrosis—present clear-cut diagnostic cues. In this case, the dermatological manifestation of unilateral flushing and sweating on the right cheek, consistently triggered by food intake, underscores the role of gustatory stimuli in the activation of aberrantly regenerated parasympathetic fibers. Dermatologists must be attuned to such presentations, particularly in patients with a prior history of herpes zoster or other cranial nerve injuries (2).

The confirmation of Lucie Frey Syndrome in our patient relied on a structured dermatologic evaluation, including a provocation test with salty foods to objectively reproduce symptoms. Advanced imaging modalities helped exclude other potential causes of facial erythema and hyperhidrosis, reaffirming the need for dermatologists to employ a multidisciplinary approach in complex cases (3).

Management in this case involved topical application of oxybutynin cream, an anticholinergic that acts by inhibiting sweat gland activation. This approach reflects a dermatologically relevant strategy for mitigating symptoms of localized hyperhidrosis.

For more sustained control, botulinum toxin injections are planned. Botulinum toxin therapy is a cornerstone in the dermatologic management of focal hyperhidrosis, offering prolonged relief by blocking acetylcholine release at the

neuromuscular junction of sweat glands (4, 5). Dermatologists should remain vigilant about these therapeutic options, tailoring treatments to individual patient needs.

This case is a rare pediatric presentation of Lucie Frey Syndrome with an infectious etiology that underscores the importance of recognizing the dermatologic sequelae of post-inflammatory autonomic neuropathies and provides insights into diagnostic techniques and treatment modalities. It also exemplifies the intricate interplay between dermatologic symptoms and underlying neuropathology, encouraging further exploration into targeted therapies for this rare condition.

4. Conclusion

This case report adds to the dermatologic literature by highlighting a rare presentation of Lucie Frey Syndrome with an infectious etiology following trigeminal herpes zoster in a pediatric patient. The unique clinical findings, diagnostic process, and management strategies outlined in this case emphasize the importance of recognizing autonomic neuropathies in the context of post-inflammatory nerve lesions. By documenting this unusual presentation, we aim to contribute to the growing understanding of this condition and inform future clinical practice

Compliance with ethical standards

Disclosure of conflict of interest

No conflict of interest to be disclosed.

Statement of informed consent

Informed consent was obtained from the participant included in the study.

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