



A Case of Frey's Syndrome Diagnosed and Followed Up Using the Quantitative Sudomotor Axon Reflex Test

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Dear Editor,

Frey's syndrome (also called focal gustatory hyperhidrosis) is a rare neurologic disorder presenting with abnormal facial sweating when consuming meals. The syndrome had been reported in up to 53% of parotidectomy patients, and may occur due to injury or inflammation of the parotid or submandibular glands, or the cervical sympathetic trunk.^{1,2} The diagnosis is traditionally made based on clinical symptoms and a starch iodine test.³ This test, while sensitive to the diagnosis of focal hyperhidrosis, does not adequately quantify sweat secretion. We report a case in which we performed the quantitative sudomotor axon reflex test (QSART) to diagnose focal hyperhidrosis, and assessed the therapeutic effect at follow-up.

A 45-year-old man visited a department of neurology for sweating in the left auriculotemporal area while consuming meals or chewing. He reported that the symptom began at approximately 30 years of age and was more severe when eating hot or sour foods. A benign branchial cleft cyst had been surgically removed from the left side of his neck in infancy. Additionally, he underwent endoscopic submucosal dissection due to early gastric cancer 1 month prior to admission, but was not taking any medication. During a neurologic examination he reported experiencing hypesthesia at the operation site since childhood. Laboratory tests including thyroid function and blood sugar, autoimmune antibody tests, electrophysiologic tests, and brain CT revealed no abnormalities.

We performed the QSART using the Q-Sweat device (WR Medical, Maplewood, MN, USA) in both auriculotemporal areas, as well as on the left arm and leg.⁴ We observed excessive sweating in the left but not the right auriculotemporal area when the patient was chewing gum (Fig. 1A). Hyperhidrosis was also observed in the left auriculotemporal area upon stimulation with acetylcholine (Fig. 1B).

The nightly application of aluminum chloride cream greatly reduced his symptoms. The total sweat volume in the left auriculotemporal area while chewing was significantly reduced at a 2-month follow-up (Fig. 1C).

Our use of the QSART confirmed the presence of excessive sweat secretion in the symptomatic auriculotemporal site induced both by either stimulation with acetylcholine or chewing. Frey's syndrome may be caused by aberrant innervation of sympathetically innervated facial sweat glands by postganglionic parasympathetic nerve fibers in damaged tissue.⁵ Therefore, mastication, which releases acetylcholine from parasympathetic nerve endings, results in sweating. The mechanism underlying Frey's syndrome in our patient was unclear, since the patient had undergone neck surgery during infancy and the symptoms occurred only when he reached his thirties. However, a case of delayed Frey's syndrome occurring 40 years after surgery has been reported.⁶ Our patient reported hypesthesia at the operation site, which might indicate the presence of similar aberrant re-innervation of the postganglionic nerve fibers near the site of his neck operation.

The QSART is a quantitative type of sudometry that is commonly used to diagnose early dia-

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betic polyneuropathy or small-fiber neuropathy.⁷ A few studies have assessed the effectiveness of treatments in patients with hyperhidrosis,^{8,9} and found the QSART to be useful in measuring the therapeutic effects. The present case has demonstrated that the QSART may also be equally useful in Frey's syndrome.

Focal hyperhidrosis could have a negative impact on social interactions, resulting in a reduced quality of life.³ It is therefore important to diagnose focal hyperhidrosis and assess the possible treatment effects in order to treat it satisfactorily, for which we believe that the QSART may be helpful.

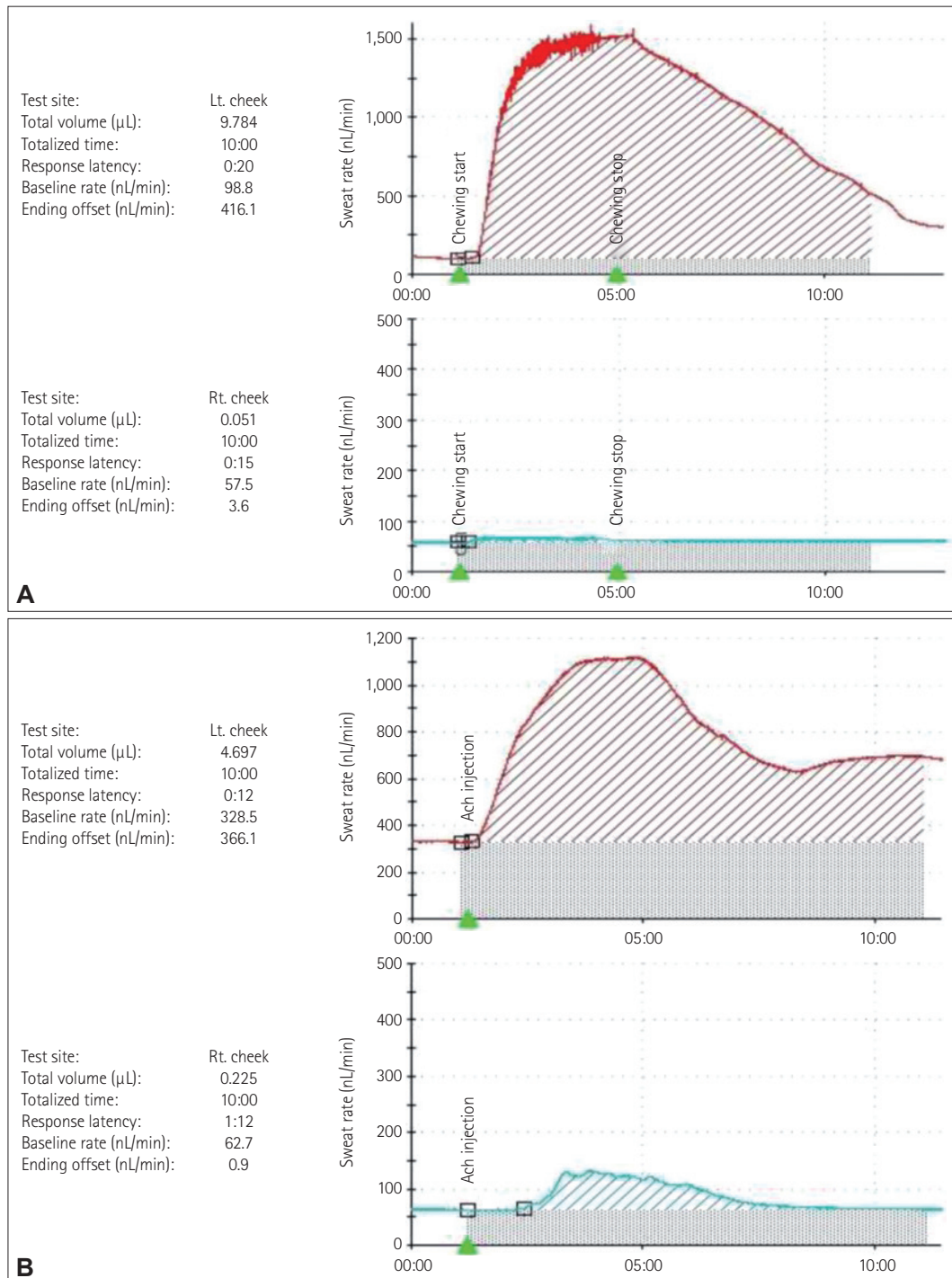


Fig. 1. The results of QSART. A: QSART findings in both auriculotemporal areas. Excessive sweating was observed on the left side after chewing gum. B: The sweat volume was larger in the left than the right auriculotemporal area. C: Sweating was significantly reduced on the left side after treatment with aluminum chloride cream at a 2-month follow-up. QSART: quantitative sudomotor axon reflex test.

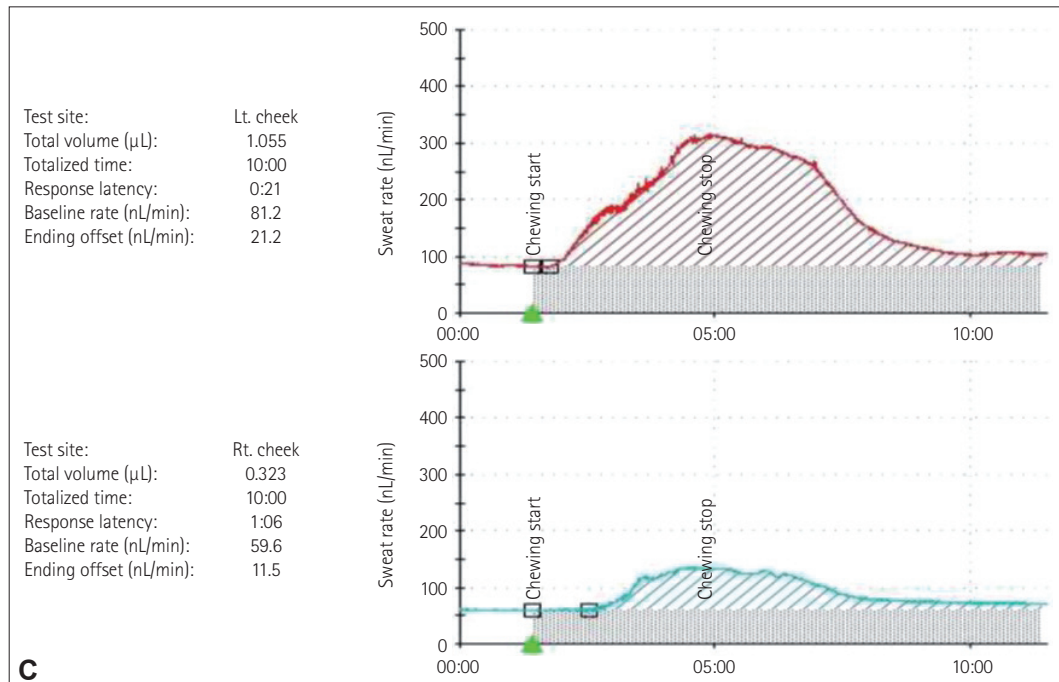


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Author Contributions

Conceptualization: Hye Lim Lee. Investigation: Jaehwan Kim, YeEun Kim, Minkyung Kim. Methodology: Jaehwan Kim, YeEun Kim, Minkyung Kim. Supervision: Hye Lim Lee. Writing—original draft: Jaehwan Kim. Writing—review & editing: Hye Lim Lee.

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Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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