Tohoku J. Exp. Med., 2006, **210**, 165-168

Case Report

Treatment of Idiopathic Gustatory Rhinorrhea by Resection of the Posterior Nasal Nerve

YAH YEE ANG, KENJI KAWANO, TATSUYA SAITO, MISATO KASAI and KATSUHISA IKEDA

Department of Otorhinolaryngology, Juntendo University School of Medicine, Tokyo, Japan

ANG, Y.Y., KAWANO, K., SAITO, T., KASAI, M. and IKEDA, K. *Treatment of Idiopathic Gustatory Rhinorrhea by Resection of the Posterior Nasal Nerve*. Tohoku J. Exp. Med., 2006, **210** (2), 165-168 —— We herein describe a case of 44-year old female who presented with a chief complaint of gustatory rhinorrhea from childhood, in which gustatory stimuli caused bilateral excessive, watery nasal secretion. No abnormality of taste acuity was observed. This disorder was presumably caused by faulty regenerated parasympathetic nerve fibers reaching the nasal mucosa or possibly, by a congenital condition. Nasal pretreatment with an anti-cholinergic drug clinically blocked the positive sugar-induced rhinorrhea, thus indicating that the gustatory rhinitis in this case was produced by foods that stimulate muscarinic receptors sensitive to atropine (probably on submucosal nasal glands). Although this syndrome can be treated prophylactically by the use of topical atropine, the patient preferred to undergo radical therapy and a resection of the posterior nasal nerve was performed through the middle meatus under endoscopic control. The resection of the nerve on both sides resulted in an almost complete inhibition of the sugar-induced rhinorrhea without serious complications. Although this disease is not life-threatening, it is socially embarrassing and troublesome to patients and surgical therapy is one of the accepted modalities. ———— gustatory rhinorrhea; posterior nasal nerve; nasal gland; muscarinic receptor

The phenomenon by which the ingestion of certain foods induces rhinorrhea is known as gustatory rhinorrhea. Gustatory rhinorrhea cases are rare, however, posttraumatic or postsurgical gustatory rhinorrhea has been occasionally reported in the literature (Stevens and Doyle 1988; Elidan and Gay 1990; Hamilton and Nettle 1990; Sadeghi et al. 1997; Langan 2004). It is thought to be caused by faulty regenerated parasympathetic nerve fibers reaching the nasal mucosa. We herein report a case with idiopathic gustatory rhinorrhea, which showed a dramatic improvement after a resection of the posterior nasal nerve through the middle meatus under endoscopic control.

CASE REPORT

A 44-year old female presented with the chief complaint of gustatory rhinorrhea related to various types of food since childhood. The symp-

Received July 18, 2006; revision accepted for publication August 7, 2006.

Correspondence: Katsuhisa Ikeda, Department of Otorhinolaryngology, 2-1-1 Hongo, Bunkyo-ku, Tokyo 113-8421, Japan.

e-mail: ike@med.juntendo.ac.jp

toms had gradually worsened over the past 25 years. She complained of no disturbance in taste or olfaction. Olfacatory stimuli showed no hypersecretion of rhinorrhea. Although she took such medication as antihistaminergic drugs, her symptoms did not improve at all.

The patient visited our clinic on November 22, 2003. No abnormality was seen on the computed tomography findings of the paranasal sinuses or allergic examinations. A provocation test by the oral administration of sugar was performed. The amount of nasal secretion was then evaluated by a filter paper placed on the patient's nares. Three g of sugar were administered, and the nasal secretion was then collected for 5 min. In order to ascertain the parasympathetic effect, oxitropium bromide (Tersigan®, Nippon Berlinger Ingelheim, Kawanishi) was topically applied to the nasal mucosa just before oral intake of sugar. The application of Tersigan® resulted in an almost complete elimination of the rhinorrhea induced by sugar ingestion at least for 3 min, thus suggesting that her gustatory rhinitis was produced by foods that stimulate atropine-inhibitable muscarinic receptors, probably in the submucosal glands (Fig. 1). Since the patient preferred to undergo radical therapy, a selective resection of the posterior nasal nerve was performed through the middle meatus under endoscopic control (Ikeda et al. 2006).

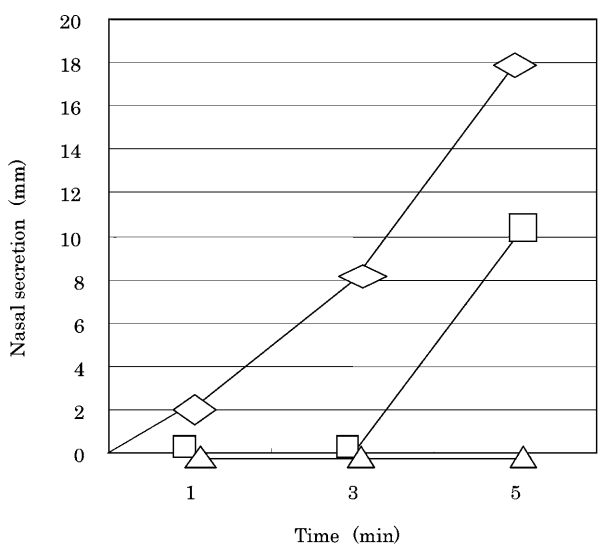

Fig. 1. Nasal secretion induced by oral intake of sugar. Nasal secretion provocated by oral administration of sugar in preoperative conditions (◇), pre-treatment with atropine sulfate oxitropium bromide in preoperative conditions (□), and postoperative conditions (△).

First, a selective resection of the posterior nasal nerves was performed using a 0 degree nasal endoscope. A 20- to 30-mm vertical incision was then made approximately 5 mm anterior to the insertion of the middle turbinate into the lateral nasal wall. The mucoperiosteal flap was then dissected posteriorly with an elevator instrument, until the crista ethomoidalis in the perpendicular plate of the palatine bone was reached. A gentle dissection above and below the crista ethomoidal then allowed for better visualization of the neurovascular bundle emerging just from the sphenopalatine foramen, which was distributed to the middle and inferior turbinates and the fontanelle. This neurovascular bundle containing the sphenopalatine vessels and the posterior nasal nerves can then be safely resected using an ultrasonic coagulator (Harmonic scalpel, Ethicon, Endo-Surgery, Cincinnati, OH, USA) to avoid any troublesome bleeding. The resected surface was covered by bony or cartilaginous fragments derived from the turbinate or nasal septum, which were fixed with fibrin glue, thereby preventing crusting, re-innervation, and hemorrhaging.

All symptoms completely disappeared just after the operation. One week after the operation, a provocation examination by sugar ingestion was performed. Nasal secretion was not induced by sugar ingestion on both sides, thereby demonstrating the clinical effectiveness of a posterior nasal nerve resection for the treatment of idiopathic gustatory rhinorrhea (Fig. 1). Effect of a posterior nasal nerve resection on idiopathic gustatory rhinorrhea lasts at least for two years.

There were no intraoperative complications. However, transient hypesthesia has been observed in both soft palates, which may have been caused by some thermal damage of the sensory nerve to the soft palate.

Discussion

We experienced a rare case of idiopathic

gustatory rhinorrhea. Posttraumatic or postsurgical gustatory rhinorrhea has been occasionally reported in the literature (Stevens and Doyle 1988; Elidan and Gay 1990; Hamilton and Nettle 1990; Sadeghi et al. 1997; Langan 2004). It is thought to be caused by faulty regenerated parasympathetic nerve fibers reaching the nasal mucosa.

Raphael et al. (1989) first reported idiopathic gustatory rhinorrhea to possibly be caused by a congenital condition, however, no substantial evidence supporting this theory was ever reported. Certain foods contain chemicals that stimulate the afferent sensory nerves in the mucosa of the mouth and oropharynx by interacting with either chemical or irritant receptors or by eliciting the release of neuropeptides from the sensory nerves (Hercer et al. 1998). However, the present case seems to show a different mechanism of hypersecretion from that reported by Raphael et al. (1989). The stimulation of the taste receptor in the oral cavity might have been involved in our case. Another possibility is olfactory or trigeminal stimuli in the nasal cavity, which will be neglected by the finding that odorants have no effect on the nasal secretion. Further study to clarify the underlying mechanism of the problem is needed.

Nasal fluid secretion is mainly due to submucosal nasal glands, with a lower contribution from an increase in the vascular and epithelial permeability. Glandular secretion is regulated by muscarinic receptor stimulation, which is mediated by parasympathetic nerves in mammals according to in vivo (Borum 1979; Gawin et al. 1997) and in-vitro studies (Sunose et al. 1994; Ikeda et al. 1995). The anti-cholinergic action of atropine dramatically inhibited the cholinergic stimulation of the secretory responses of guinea-pig nasal glands in both guinea pigs and humans in our laboratory (Sunose et al. 1994; Wu et al. 1994; Ikeda et al. 1995; Furukawa et al. 1996). These findings convincingly support that an interruption of the parasympathetic stimulation suppresses the secretory events of the nasal gland. In fact, Konno et al. (1987) demonstrated that a vidian neurectomy resulted in a complete block of antigen-induced hyperresponsiveness of the nasal mucosa and the resulting nasal hypersecretion. In the present case, nasal pretreatment with an anti-cholinergic drug clinically blocked the sugar-induced rhinorrhea, thus indicating that the gustatory rhinitis in this case was produced by foods that stimulate atropine-inhibitable muscarinic receptors (probably on the submucosal glands). However, the fact that pre-treatment of oxitropium bromide resulted in incomplete inhibition of the nasal secretion in 5 min suggests that the gustatory rhinorrhea is partly brought about by an increase in the vascular and epithelial permeability.

We performed a resection of the posterior nasal nerve innervating the nasal submucosal gland in order to reduce the hyperrhinorrhea. The symptomatic improvement of hyperrhinorrhea and the inhibition of sugar-induced hyperrhinorrhea were thus obtained. The surgical resection of the posterior nasal nerve may be an effective treatment for gustatory rhinorrhea.

References

Borum, P. (1979) Nasal methacholine challenge: a test for the measurement of nasal reactivity. *J. Allergy Clin. Immunol.,* **63**, 253-257.

Elidan, J. & Gay, I. (1991) Posttraumatic gustatory rhinorrhea. *Ear Nose Throat J.*, **70**, 184-185.

Furukawa, M., Yamaya, M., Ikeda, K., Oshima, T., Suzuki, T., Sasaki, H. & Takasaka, T. (1996) Culture and characterization of human nasal gland cells. *Am. J. Physiol.,* **271**, L593-L600.

Garwin, A.Z., Emery, B.F., Baraniuk, J.N. & Kaliner, M.A. (1991) Nasal glandular secretory response to cholinergic stimulation in human and guinea pigs. *J. Appl. Physiol.,* **71**, 2460-2468.

Hamilton, R.B. & Nettle, W.J. (1990) Gustatory rhinorrhea after radical parotidectomy. *Scand. J. Plast. Reconstr. Surg. Surg.,* **24**, 163-166.

Hercer, M., Shnarch, A., Benoliel, R. & Sharav, Y. (1998) Trigeminal neuralgic-type pain and vascular-type headache to gustatory stimulus. *Headache*, **38**, 129-131.

Ikeda, K., Ishigaki, M., Wu, D., Sunose, H., Suzuki, M., Ishitani, K. & Takasaka, T. (1995) Intracellular Ca^{2+} responses induced by acetylcholine in the submucosal nasal gland acinar cells in guinea pigs. *Am. J. Physiol.,* **268**, L361-L367.

Ikeda, K., Oshima, T., Suzuki, M., Suzuki, H. & Shimomura, A. (2006) Functional inferior turbinosurgery (FITS) for the treatment of resistant chronic rhinitis. *Acta Otolaryngol.,* **126**, 739-745.

Konno, A., Terada, N., Okamoto, Y. & Togawa, K. (1987) The role of chemical mediators and mucosal hypereactivity in nasal hypersecretion in nasal allergy. *J. Allergy Clin. Immunol.,* **79**, 620-626.

Langan, M.L. (2004) Gustatory rhinorrhea as a complication of oral surgery. *J. Am. Geriatr. Soc.,* **52**, 1786-1787.

Rapahel, G., Raphael, M.H. & Kaliner, M. (1989) Gustatory Rhinitis: a syndrome of food-induced rhinorrhea. *J. Allergy Clin. Immunol.,* **83**, 110-115.

Sadeghi, H.M., Siciliano, S. & Reychler, H. (1997) Gustatory rhinorrhea after maxillectomy. Two case reports are considerations on etiology and pathophysiology. *Int J. Oral. Maxillofac. Surg.,* **26**, 124-126.

Stevens, H.E. & Doyle, P.J. (1988) Bilateral gustatory rhinorrhea. following bilateral parotidectomy a case report. *J. Otolaryngol.,* **17**, 191-193.

Sunose, H., Zhang, W., Ishigaki, M., Katori, Y., Suzuki, M., Ikeda, K., Takasaka, T., Saito, Y. & Nishiyama, A. (1994) Isolation of acini from nasal glands of the guinea-pig. *Acta Physiol. Scand.,* **151**, 377-384.

Wu, Z., Sunose, H., Ikeda, K., Ishigaki, M. & Takasaka, T. (1994) Ionic currents evoked by acetylcholine in isolated acinar cells of the guinea pig nasal gland. *Biochem. Biophys. Res. Comm.,* **202**, 307-313.