A case of gustatory rhinorrhoea*

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SUMMARY *The authors describe a case of gustatory rhinorrhoea that appeared one year after skull trauma with delayed facial palsy. Traumatic interruption and abnormal regrowth of salivary parasympathetic fibers is hypothesized. In order to explain the pathogenesis of this syndrome an anatomical review of the transpetrosal nerves is included. A review of the literature is also presented.*

Keywords: gustatory rhinorrhoea, anomalous gustatory reflex, autonomic nervous system, transpetrosal nerves, skull trauma

INTRODUCTION

Anomalous gustatory reflex syndromes are congenital or acquired diseases in which gustatory input is followed by a reflex different from salivation, determined by the autonomic nervous system. The most common is Frey's syndrome, in which the reflex provokes sweating of the laterocervical region, usually determined by the sympathetic system. Other syndromes are "crocodile tears", gustatory rhinorrhoea and gustatory otorrhoea, in which the gustatory input provokes phenomena other than salivation because the efferent reflex arc may take different pathways (Alexon et al., 1963; Readleaf et al., 1993; Berrettini et al., 1994).

Our patient presented an annoying periprandial rhinorrhoea that appeared about one year after a skull trauma with delayed facial nerve palsy. We classified this case as an anomalous gustatory reflex syndrome. We also present an anatomical review of the transpetrosal nerves with their anastomoses in order to explain the anomalous gustatory reflex syndrome.

CASE REPORT

In 1992 a 34-year-old physician had a left skull trauma in consequence of a car accident. Three days later a left facial nerve paralysis appeared. A CT scan of the skull and temporal bone revealed a transversal fracture of the petrosal bone with a haemotympanum. Surgical therapy was not performed, because the palsy appeared three days after the trauma, so that resection of the nerve could be excluded. Medical therapy included phenobarbital (100 mg/day), dexamethasone (8 mg/day), and co-trimoxazole (160 mg trimethoprim, 800 mg sulfamethoxazole; twice daily for the first week). Afterwards chemotherapy was interrupted and the patient continued to take dexamethasone (4 mg/day) for 10 days and phenobarbital (100 mg/day) until now. In a few months the paralysis recovered with residual synechias. One year later the patient returned for an annoying left rhinorrhoea that appeared during meals. The clinical examination revealed left facial synechias, left facial rosacea and a slight hearing loss at the left. Furthermore, he complained of hypogeusia of the left part of the tongue. The rhinorrhoea did not change depending on body position and did not appear during coughing or defaecation. Nevertheless, in order to exclude the presence of rhinocerebral fluid, a fronto-ethmoidal thin-slice CT scan was made and the presence of glucose in the nasal liquid was tested, but both gave negative results. Since the patient described the occurrence of rhinorrhoea during meals, an abnormal gustatory re-innervation syndrome was suspected. In order to confirm the diagnosis, the tip of the patient's tongue was stimulated with lemon juice and then the beginning of a rhinorrhoea from the left nostril was verified. Rhinomanometry showed an increased nasal resistance at the left after gustatory input. Mucociliary transport was been not studied. "Crocodile tears" as a possible cause of rhinorrhoea was excluded by the Schirmer test. Therefore, the final diagnosis was: "monolateral post-traumatic gustatory rhinorrhoea from abnormal parasympathetic re-innervation".

DISCUSSION

The ascending gustatory reflex arc is formed by fibers sensitive to gustatory, thermic and tactile inputs presented to the mouth and tongue receptors. Fibers originating from two-thirds of the fore-tongue run along the lingual nerve and the chorda tympani (Figure 1, No. 11) to the geniculate ganglion and they arrive at the *nucleus solitarius* (NS) through Wrisberg's nerve. The NS

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has connections with all salivary nuclei and represents an important center for all gustatory fibers: (1) fibers from the soft palate arrive at the NS through the *nervi palatini*, the sphenopalatine ganglion, the Vidian nerve (Figure 1, No. 3), the *nervus petrosus superfacialis major* (Figure 1, No. 2), the geniculate ganglion (GG) and Wrisberg's nerve; (2) fibers from one-third of the posterior aspect of the tongue go through the maxillary ramus of the glossopharyngeal nerve and through the glossopharyngeal nerve itself to the NS.

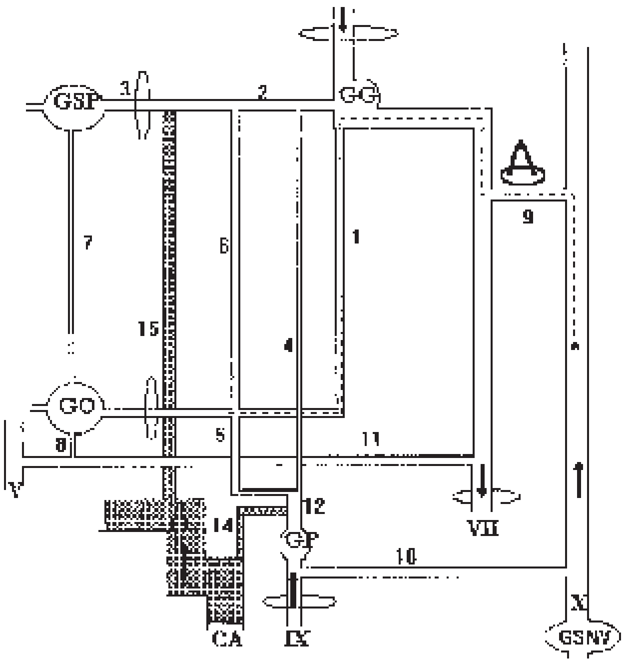

Figure 1. Schematic diagram of the gustatory reflex arc (CA: carotid artery; IX: nervus glossopharyngeus; VII: nervus facialis; X: nervus vagus; V: nervus lingualis; GP: ganglion petrosus; GG: ganglion geniculatus; GSP: ganglion sphenopalatinus; OG: ganglion oticus; GSNV: ganglion superior nervi vagi; 1: nervus petrosus superfacialis minor; 2: nervus petrosus superfacialis major; 3: nervus vidianus; 4: nervus petrosus profundus major; 5: nervus petrosus profundus minor; 6: anastomosis between nervus petrosus superfacialis major and nervus petrosus profundus minor; 7: nervus sphenoidalis internus (sphenoidal internal nerve); 8: anastomosis between ganglion oticus and chorda tympani; 9: terminal branch of the ramus auricularis nervi vagi; 10: fibers from ganglion petrosus to nervus vagus; 11: chorda tympani; 12: Jacobson's nerve; 13: carotic plexus; 14 and 15: sympathetic fibers from carotic plexus to petrosal nerves; *: fibers passing from nervus vagus to nervus petrosus profundus major through nervus petrosus superfacialis minor).

Pre-ganglionic parasympathetic fibers coming from the inferior salivary nucleus go through the glossopharyngeal nerve, Jacobson's nerve (Figure 1, No. 12) and the *nervus petrosus profundus minor* (Figure 1, No. 5). They arrive at the otic ganglion (OG), which is located under the *foramen ovale*, where they meet with post-ganglionic fibers reaching the parotid gland through the auriculotemporal nerve.

Superior salivary and lacrimal nucleus fibers go through Wrisberg's nerve. Fibers going to the lacrimal glands and the nasal mucosa go through the *nervus petrosus superfacialis major* (PSM; Figure 1, No. 2) and the Vidian nerve (Figure 1, No. 3), then they reach the sphenopalatine ganglion (GSP).

The GSP is located in front of the Vidian canal in the pterygopalatine fossa. Post-ganglionic fibers go to the lacrimal gland, through an efferent ramus, join the maxillary nerve and then its *ramus zygomaticus*. These fibers are likely to follow the *ramus zygomaticus-temporalis* and, through a *ramus anastomoticus*, the lacrimal nerve and reach the glandular tissue (Gray, 1980). Fibers going to the nasal mucosa go through the *nervus palatinus anterior* and the nasal rami. On its way in the *canalis palatinus major* the *nervus palatinus anterior* issues the inferior nasal nerves which innervate the inferior turbinate, the superior and the middle meatal mucosa, passing through the *lamina perpendicularis ossis palatini foramina*. The nasal nerves reach the nasal fossa through the pterygopalatine foramen, then they divide in the medial and the lateral posterosuperior nasal nerves and innervate the middle and the superior turbinates, the superior part of the nose, the superior meatus and the ethmoid cells (Legent et al., 1979; Gray, 1980). Fibers going to the submandibular and the sublingual glands leave Wrisberg's nerve and reach the submandibular ganglion through the chorda tympani (Figure 1, No. 11).

An accurate study of the temporal bone nerves reveals a complicated and variable anatomy because of many anastomosis between Wrisberg's nerve and the glossopharyngeal nerve due to the common embryonic origin of the geniculate ganglion (GG) and glossopharyngeal ganglia. Some authors find a connection between the geniculate and glossopharyngeal ganglia in the 16.5-mm human embryo, which was supposed to be Jacobson's plexus precursor (Gasser, 1966; Vidic et al., 1967; Vidic, 1969).

The terminal branches of Jacobson's nerve (Figure 1, No. 12) are the *nervus petrosus profundus major* (PPM; Figure 1, No. 4) and the *nervus petrosus profundus minor* (PPm; Figure 1, No. 5; cf. Bessede et al., 1983).

The nervus facialis passing in its temporal canal issues the PSM (Figure 1, No. 2), the *nervus petrosus superfacialis minor* (PSm; Figure 1, No. 1), the *nervus stapedius* and the chorda tympani (Figure 1, No. 11).

The PPm (Figure 1, No. 5) in the middle cranial fossa receives the PSm (Figure 1, No. 1) and this recurring anastomoses represents a possible way for anomalous re-innervation. The PSm (Figure 1, No. 1) is a little branch basically formed by small myelinated fibers (numbering 50-300) that can depart either from the second portion of the facial nerve, from the CG or from the first part of the PSM (Figure 1, No. 2). Sometimes, the PPm seems to be linked with the second portion of the facial nerve or with the GG, so that the PSm is not present. The PSm-fibers (Figure 1; asterisk) arise from a terminal branch of the *ramus auricularis nervi vagi* passing through the facial canal. Fibers from this branch of the *nervus vagus* likely pass into the PSM (Figure 1, No. 2) too. It may that the parasympathetic preganglionic fibers innervate the parotid gland. It is necessary to stress that the *ramus auricularis nervi vagi* receives fibers coming from the petrosal ganglion of the glossopharyngeal nerve just after its arising from the *ganglion superior nervi vagi* (Figure 1, No. 10 [Vidic et al., 1967; Vidic, 1969; Gray, 1980]).

The PPM (Figure 1, No. 4) arrives onto the *tegmen tympani* where it reaches the PSM (Figure 1, No. 2) coming from the facial nerve. Then they receive sympathetic fibers (Figure 1, No.

15) from the carotic plexus forming the Vidian nerve (Figure 1, No. 3) that, through the Vidian canal, arrives at the GSP. The PPM (Figure 1, No. 4) is formed both by pre-ganglionic parasympathetic fibers, coming from the *nucleus salivatorius inferior*, and by sympathetic fibers arising from the carotic plexus through the *nervus carotico-tympanicus* (Figure 1, No. 14 [Gray, 1980; Bessede, 1983]).

Brzezinnski (1963) noticed the existence of an anastomosis (Figure 1, No. 6) formed by sensory fibers, which connects the PSM (Figure 1, No. 2) with either the PSm (Figure 1, No. 1) or the PPm (Figure 1, No. 5 [Stevens et al., 1988; Elidan et al., 1990]).

GSP and OG are connected by the sphenoidal internal nerve (Figure 1, No. 7) which is not always present (Gray, 1980; Bessede et al., 1983; Elidan et al., 1990); also, an inconstant anastomosis (Figure 1, No. 8) between the OG and the chorda tympani (Figure 1, No. 11) has been described (Gray, 1980).

In conclusion, there are at least five possible anastomoses between the parasympathetic fibers of the facial and glossopharyngeal nerves. They are all potential ways for anomalous parasympathetic re-innervations. Gustatory rhinorrhoea has been described in only four patients, three following parotidectomy (Boddie et al., 1976; Stevens et al., 1988; Hamilton et al., 1990) and one after skull trauma (Elidan et al., 1990).

Boddie et al. (1976) reported the first case of gustatory rhinorrhoea after a parotidectomy. They hypothesized that parasympathetic salivary fibers innervating the parotid gland, normally passing through the PPm (Figure 1, No. 5), degenerated after the parotidectomy and regrew abnormally going into the Vidian nerve (Figure 1, No. 3) through connections (Figure 1, No. 6) between this nerve and the PPm.

Stevens and Doyle (1988) described a case of bilateral gustatory rhinorrhoea after a bilateral parotidectomy. They proposed as abnormal re-innervation ways both the one proposed by Boddie et al. (1976), and also that fibers directed to the parotid gland from the facial nerve and chorda tympani (Figure 1, No. 11) can pass directly from the OG to the PSM (Figure 1, No. 2), the Vidian nerve (Figure 1, No. 3) or to the GSP through the sphenoidal internal nerve (Figure 1, No. 7). The patient recovered after resection of the Vidian nerves. This case is very interesting because this complication of parotidectomy is extremely rare and it is improbable that it can happen twice in the same patient. It may mean that this abnormal re-innervation happens when a particular anatomical condition exists and not casually.

The only case of post-traumatic gustatory rhinorrhoea was described by Elidan and Gay (1990) in a patient who had a skull trauma. They presented an extensive anatomical review proposing the same hypotheses furnished by Stevens and Doyle (1988).

In case of traumatism it is possible to hypothesize the interruption of both fibers going to the salivary glands and fibers going to nasal mucosa with consequent substitution of the ones by the others. On the other hand, it is difficult to explain gustatory rhinorrhoea after parotidectomy. In this case, parasympathetic fibers originally directed to the parotid gland have to substitute normal fibers directed to the nasal mucosa or have to place themselves side by side with them.

We think that, in our cases the fracture interrupted Wrisberg's nerve salivary parasympathetic fibers going to the sublingual and submandibular glands through the chorda tympani (Figure 1, No. 11), as evidenced by the left disgeusia of our patient.

The fracture also interrupted sympathetic fibers going to the zygomatic skin arterioles passing through the PSM (Figure 1, No. 2) and fibers directed to the nasal mucosa passing through the same nerve. The occurence of rosacea may be due to the interruption of sympathetic vasomotor fibers from the superior cervical ganglion directed to the zygomatic skin region. Those fibers (Figure 1, No. 15) come from the superior cervical ganglion and go through the carotic plexus (Figure 1, No. 13), the PPM (Figure 1, No. 2), the Vidian nerve (Figure 1, No. 3), the GSP and the mandibular nerve. In this case the rosacea would be a chalasia, that is a hypotonia of arteriolar muscular fibers due to a sympathetic denervation. Then, the parasympathetic fibers running in Wrisberg's nerve and originally directed to the chorda tympani (Figure 1, No. 11) re-innervate the PSM (Figure 1, No. 2) going to the GSP and then to the nasal mucosa.

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