Phantosmia and Dysgeusia following Endoscopic Transcribriform Approaches to Olfactory Groove Meningiomas

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Abstract

The endoscopic, endonasal transcribriform approach (EETA) is an important technique used to directly access the anterior skull base and is increasingly being used in the management of olfactory groove meningiomas (OGMs). As this approach requires removal of the cribriform plate and olfactory epithelium en route to the tumor, patients are anosmic postoperatively. Here, we report the development of phantosmia and dysgeusia in two patients who underwent EETAs for OGMs, which has not yet been reported in the literature. We hypothesize that phantosmia and dysgeusia may result from aberrant neuronal signals or misinterpretation centrally from the remaining distal portions of the olfactory and taste pathways. Since EETAs are newer than traditional open craniotomy-based techniques, reporting these outcomes will be important to appropriately counsel patients preoperatively.

Keywords

- ▶ olfactory groove meningioma
- ▶ transcribriform
- ▶ phantosmia
- ▶ dysgeusia
- ▶ anterior skull base

Introduction

Olfactory groove meningiomas (OGMs) comprise an important subset of meningiomas due to their location along the anterior skull base and association with critical neurovascular structures. Surgical options for removal of these tumors include both traditional open and newer endoscopic, endonasal transcribriform approaches (EETAs). As experience with EETAs for resection of anterior skull base meningiomas increases, evaluation of outcomes is necessary to best understand relative advantages and disadvantages between these two approaches. Here, we discuss the uncommon development of phantosmia and dysgeusia that can result from transcribriform approaches in the treatment of OGMs in two patients.

OGMs arise from the arachnoid overlying the cribriform plate of the ethmoid bone and account for approximately 10% of all intracranial meningiomas.^{1,2} OGMs are most often benign tumors that can be cured surgically if a gross total resection can be achieved. Their relatively slow growth rate, and thus the development of compensatory changes in surrounding tissue, allows these tumors to grow large before causing symptoms.¹ Symptoms correlate with tumor size and proximity to critical neurovascular structures, which can cause olfactory and optic nerve changes by mass effect, behavioral changes by causing frontal lobe dysfunction, disruption of electrical activity of neural tissue leading to cortical irritation and/or frank seizure activity, and erosion of the anterior skull base and paranasal areas potentially leading to cerebrospinal fluid (CSF) leak.^{2,3}

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Both traditional craniotomy and newer EETAs are used to surgically treat OGM. The surgical corridor provided by each technique is guided by providing access not only to the tumor but also to the dural tail and its vascular supply, which usually arises from the ethmoidal arteries along the medial anterior fossa floor. When an OGM is approached via open craniotomy, which can include pterional, bifrontal transbasal, or supraorbital approaches,^{4–6} a bone flap is created to gain access around the periphery of the tumor and the vasculature along the skull base. EETAs provide direct access inferiorly to the tumor and the ethmoidal vessels with a panoramic view.^{7–9} Although open craniotomy techniques have an advantage of having relatively higher rates of gross total resection and lower rates of CSF leak, EETAs are less invasive, allow early identification of feeding vessels, and allow removal of potentially hyperostotic ethmoidal bone en route to the OGM.¹⁰ As experience with EETAs accumulate, reporting adaptations in technique designed at mitigating complications and increasing efficacy is critical for enhancing patient outcomes.^{7,11}

Abnormalities in the sense of smell are common in patients with OGM.¹ Specific olfactory symptoms may range from anosmia/hyposmia to distorted smell perception (dysosmia) even in the absence of a smell stimulus (phantosmia). No current reports of abnormal taste perception associated with anterior fossa pathology have been reported to our knowledge. To increase knowledge of potential adverse side effects in patients with OGMs, we report olfactory and

gustatory symptoms including phantosmia and dysgeusia (abnormal taste perception) in two patients who underwent EETA for OGM. The mechanisms by which abnormal special sensory phenomena develop are not clear and currently few medical treatments are available.^{12,13} Thus, these potential effects may be useful in the preoperative counseling of patients in whom EETAs are being considered.

Patient 1

Perioperative Course

A 35-year-old woman with a history of intermittent dysphagia for 7 years presented with headache and hyposmia. Sinus computed tomography (CT) showed a left maxillary retention cyst and magnetic resonance imaging (MRI) of the brain revealed a 3.2-cm OGM, with superior displacement of the orbitofrontal and frontopolar arteries (►Fig. 1). The patient preferred a less invasive surgical approach and was scheduled for an EETA. On the planned day of surgery, she presented with rhinorrhea and pharyngitis, and thus only an endonasal exploration, sphenoidotomy, and ethmoidectomy were performed, but the intracranial portion of the surgery was postponed due to the increased risk of infection. Two weeks later, rhinoscopy showed no evidence of residual infection. Six weeks later, she underwent EETA with stereotactic guidance in which the entire cribriform plate was removed. The anterior border of bony resection approached the crista galli

Fig. 1 Magnetic resonance imaging of the brain for patient 1. (a–c) Preoperative axial, coronal, and sagittal T1 postcontrast images shows a 3.2-cm olfactory groove meningioma with superior displacement of the orbitofrontal and frontopolar arteries. (d–f) Postoperative T1 postcontrast images shows gross total resection of the meningioma.

and the posterior border approached the edge of the planum sphenoidale. The dura and tumor were removed while carefully avoiding the olfactory bulbs, and then a pedicled nasoseptal flap was used for closure of the skull base. Final pathologic analysis revealed a World Health Organization (WHO) grade I meningioma. Immediate postoperative period was uneventful except for expected anosmia.

Postoperative Course

At 4 months postoperatively, she presented with a persistent, noxious, metallic, or bleach-like taste in her mouth that was exacerbated by drinking. She was able to distinguish sweet, salty, bitter, and sour tastes. She had persistent anosmia. The remainder of her neurologic exam was normal. Provocative maneuvers for CSF leak were negative. CT and MRI were notable for only postoperative changes, with no evidence of ischemia. A 2-week course of amoxicillin-clavulanate was prescribed to treat any possible subacute infection without improvement. At 7 months, she continued to have anosmia. Her dysgeusia had evolved from a bleach taste to a persistent moldy or dirty sensation in the mouth. Nasal endoscopy revealed a healthy septal mucosal flap extending over the majority of the skull base. At 10 months, she presented with left-sided facial weakness and she was treated with a 7-day course of valacyclovir and prednisone for possible Bell's palsy.

CT scan of the head was unremarkable. Endocrinologic evaluation was normal. Her facial weakness ultimately resolved but her dysgeusia persisted.

Patient 2

Perioperative Course

A 40-year-old woman with a history of left optic nerve sheath meningioma treated with fractionated stereotactic radiotherapy presented with frontal headaches and preexisting left visual loss. MRI showed a 1.1 cm, partially calcified, enhancing extra-axial lesion along the left olfactory groove consistent with a meningioma (►Fig. 2). Examination of the nasopharynx, oropharynx, oral cavity, and neck was normal, and her smell was intact. She underwent an EETA for her OGM under stereotactic guidance. The inferior portions of the superior turbinates were resected, sphenoidotomies were created, and a left nasoseptal flap was harvested. The cribriform plate, left ethmoid roof, and crista galli were drilled to expose dura from the left orbit to the midline to expose the margins of the tumor. The tumor involved the falx cerebri and was partially calcified. A Simpson grade I resection was performed, while carefully preserving the olfactory bulbs. Allograft and a pedicled nasoseptal flap were used to complete the skull base repair. Final pathologic analysis revealed a

Fig. 2 Magnetic resonance imaging of the brain for patient 2. (a, b) Preoperative axial and coronal T1 postcontrast images shows a 1.1-cm left olfactory groove meningioma. (c, d) Postoperative T1 postcontrast images shows gross total resection was achieved.

WHO grade I meningioma. Her immediate postoperative course was unremarkable aside from the expected anosmia, and she was discharged home on postoperative day 3.

Postoperative Course

Four weeks postoperatively, she reported malaise, nasal congestion, facial pressure, and postnasal drainage. Endo-nasal examination revealed sinonasal crusting and increased mucus. The symptoms resolved with saline irrigations and a 2-week course of amoxicillin/clavulanic acid. Six weeks after surgery, she started experiencing constant sensations of an unpleasant sweet-smelling phantosmia and “foul” distorted taste of food (aliageusia). There was no headache or nasal drainage and her neurologic exam was otherwise unchanged. Endonasal examination showed a healthy nasal graft and a healed graft harvest site. Her phantosmia and dysgeusia have persisted at her most recent clinic appointment 5 months postoperatively.

Discussion

EETAs constitute new techniques that are increasingly being used to resect OGMs in selected patients. The relative advantages of this technique over open craniotomy approaches are that EETAs are less invasive, obviate the need for brain retraction, and allow early identification and coagulation of the tumor blood supply and direct access to the dural tail and any hyperostotic bone, facilitating a complete Simpson grade I resection. Relative disadvantages are the increased risk of CSF leak compared with open craniotomy, the increased rate of subtotal resection, and the need for specialty neuroendoscopy training.^{7,10} As experience with new techniques accumulates, reporting complications will be important to design operative strategies to minimize complications and enhance patient outcome. These two patients illustrate the potential for abnormalities of smell and taste that can arise postoperatively in patients with OGMs after EETAs (►Table 1). Our first patient initially presented with hyposmia that progressed to anosmia, as expected after an EETA, but also subsequently

developed dysgeusia postoperatively. Although she developed Bell’s palsy which can result in taste disturbances, she had these symptoms prior to the facial palsy and she had preserved ability to distinguish four basic tastes—salty, sweet, sour, and bitter—suggesting that the neurological pathways for taste were intact. Our second patient did not report any symptoms related to smell or taste preoperatively; however, she developed significant unpleasant phantosmia and dysgeusia postoperatively.

Anosmia is common in patients with OGM preoperatively as well as after surgery.¹ In a large series of patients surgically treated for OGM, anosmia was present in almost 60% of patients preoperatively.⁶ Anosmia in patients with OGM can be caused by damage to olfactory epithelium, nerves, bulbs, or tracts, from tumor invasion, or from surgical excision of the tumor. The olfactory bulbs may be adherent to the tumor making their preservation difficult. Patients with olfactory loss before surgery seldom recover the sense of smell after surgery. If olfaction is intact preoperatively, however, a craniotomy approach offers a reasonable chance of preserving olfaction.¹⁴ EETAs allow direct visualization of the olfactory bulbs and tracts but require removal of the middle and superior turbinates as well as a large septectomy resulting in resection of olfactory epithelium and transection of olfactory axons as they pass through the cribriform plate and synapse onto the olfactory bulbs. A recent review of the literature did not reveal a significant difference in the incidence of postoperative anosmia between open and endoscopic techniques of OGM resection.¹⁵

Smell and taste are intimately related. Taste itself is an oropharyngeal chemical sense activated by interaction of chemicals in food or drink with cation channels or G-protein coupled receptors on the surface of epithelial taste receptor cells in taste buds. Each taste bud contains approximately 50 to 100 tightly packed taste receptor cells in addition to basal progenitor cells and supporting cells. Clusters of taste buds are located in taste papillae on the tongue and adjacent areas in the mouth as well as the oropharynx. Five specific tastes—sweet, sour, salty, bitter, and umami—have been described. The impulses from taste receptor cells are transmitted to the brain via branches of the facial, glossopharyngeal, and vagus nerves geographically arranged on the tongue, oral cavity, and posterior oropharynx.^{16,17} Most patients who complain of apparent dysgeusia have synchronous disorders of olfaction. In one study, 433 of 586 (74%) patients presenting with anosmia complained of decreased taste, although formal testing revealed loss of taste in less than 4%.¹⁸ Moreover, patients presenting only with disorders of taste were three times more likely to have olfactory deficit than a gustatory deficit.¹⁸

Flavor is a broader sensation incorporating taste, smell, texture, and temperature.^{12,13} Altered sense of smell with preserved neurological pathways for taste may lead to dysgeusia due to reduced ability to appreciate flavor. Smell is an important component of flavor because the human nose is able to detect more than 1 trillion different smells, which is considered more complex than the other special senses.¹⁹ Abrupt anosmia may seriously impair the ability to detect flavor but with time the body can often learn to compensate

Table 1 Patient demographics and meningioma characteristics

	Patient 1	Patient 2
Age (y)	35	40
Gender	Female	Female
Presentation	Headache and hyposmia	Frontal headache
Postoperative symptoms	Dysgeusia	Dysgeusia and phantosmia
Tumor location	Olfactory groove/planum sphenoidale	Left olfactory groove
Tumor size (cm)	3.2	1.1
WHO grade	Grade I	Grade I
Ki-67 index (%)	1.2	2.5

Abbreviation: WHO, World Health Organization.

for loss of smell by emphasizing other components of flavor such as taste, texture, and temperature. Thus, patients with Kallman's syndrome—an X-linked congenital syndrome characterized by hypogonadism and congenital anosmia despite intact taste pathways—rarely complain of decreased flavor perception.²⁰ Given the intact ability for true taste after surgery, the presented cases likely developed complaints of altered “taste” as an effect of distortions of smell on flavor. An underlying phantosmia is more likely the cause in this situation for manifestations of dysgeusia. Postoperative phantosmia and dysgeusia after OGM surgery may be expected to diminish with time possibly from central suppression of sensory stimuli,²¹ development of alternative mechanisms of flavor detection, or potentially from regrowth of olfactory neurons from any remaining portions of the olfactory epithelium projecting to intact olfactory bulbs.²¹

We were unable to find a previous report of phantosmia and dysgeusia from OGM in the literature; our patients appear to be the first such reported cases. Reduction or distortion of smell and taste significantly impacts quality of life,^{22,23} is associated with many chronic disease processes,²⁴ and may increase risk of exposure to environmental hazards that are normally detected by normal olfaction.²⁵ Henkin and Hoyer²⁶ and Hoyer et al²⁷ described 17 patients with paranasal malignancies requiring excision of the cribriform plate and removal of olfactory epithelium. Immediately, postoperatively, most had complete anosmia. Subsequently over 2 weeks to 6 months, several patients developed some degree of smell. It is hypothesized that loss of olfaction causes new dysplastic pathways that distort smell and taste perception. Mechanistic studies have shown that specific regions of gustatory and olfactory cortex are activated during phantom smell and taste perception using functional MRI, which can be inhibited using thioridazine or haloperidol.²⁸ Furthermore, Levy and Henkin²⁹ and Henkin et al³⁰ have reported reduced gamma-aminobutyric acid (GABA) levels in functional MRI scans of nonsurgical patients with phantosmia and phantageusia, suggesting a potential avenue for therapeutic intervention with GABA receptor agonists.³¹

Proposed treatments for parosmia and/or dysgeusia include zinc supplementation,³² nasal irrigation with saline, antidepressant medications,^{33,34} treatment of associated conditions such as allergic or bacterial rhinitis and gastroesophageal reflux, GABAergic medications,³¹ and surgical resection of olfactory bulbs or olfactory epithelium.^{35,36} Clonazepam has been found to be particularly effective at reducing burning sensation of mouth.³⁷ In a follow-up study of 48 patients with dysgeusia secondary to a variety of non-neurosurgical causes such as periodontal or paranasal sinus disease, gastroesophageal reflux disease, menopause, and vitamin B12 deficiency, resolution of dysgeusia occurred in 63% with an average time of resolution of 10 months.³³ In a clinical trial with sham controls, transcranial magnetic stimulation was reported to exert positive effects against phantosmia and dysgeusia based on prior functional MRI studies,^{28,30,38} but further study is probably required before widespread adoption of this technology.^{38,39} In general, therapy for patients with either phantosmia or dysgeusia in any context merits continued exploration and innovation.

A thorough understanding of potential adverse effects from EETAs is necessary as our experience with this technique grows. Here, we present two cases of phantosmia and dysgeusia that developed after EETAs for resection of OGMs. Patients with OGMs selecting EETAs should be formally tested for smell and taste before surgery, and patients and neurosurgeons should be aware of the postoperative risk of phantosmia and dysgeusia.

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