



Case report

Endoscopic Vidian neurectomy for treating postcoital unilateral hydrorhinorrhea: A case report and literature review

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ABSTRACT

Introduction and importance: Hydrorhinorrhea in response to sexual intercourse and orgasm (*honeymoon rhinitis*) is an underreported clinical presentation producing physical and behavior discomfort. Vidian neurectomy has been proposed for treating this condition when pharmacological treatment fails.

Case presentation: We present a case of honeymoon rhinitis consisting of a 48 years-old male patient with left unilateral nasal discharge mainly present during sexual intercourse and orgasm. Exploratory nasal endoscopy, CT and MRI were normal. Beta-trace test and all allergy tests were negative. The response to antihistamines and corticosteroids had been negative. A Vidian endoscopic neurectomy was carried out.

Clinical discussion: Using Vidian neurectomy, a complete sympathetic-parasympathetic denervation of the nasal mucosa was achieved. After a follow-up of 15 days, the patient stated that his symptoms had improved. Nasal obstruction, sneezing, and rhinorrhea had decreased without showing symptoms either in the morning or during sexual intercourse. After two years of follow-up, the symptoms were still resolved without any complaints related to his preoperative "honeymoon rhinitis."

Anatomical structural relationships between the activation of the autonomic nervous system in the pelvic region and the stimulation of sympathetic-parasympathetic neurons in the nasal mucosa is unknown. However, the use of Vidian neurectomy for treating these cases reveals they may be mediated by a possible disbalance of the autonomic activity.

Conclusion: This case report gives background on the autonomic innervation of the nasal mucosa and how its imbalance causes a clinical condition that we suggest it could be solved by the Vidian neurectomy when other therapeutic measures fail.

1. Introduction

The relationship between the nose and the reproductive organs was described by Watson in 1875 [1] and Mackenzie in 1884 [2]. They referred to the phenomena that connected changes in the nasal mucosa to pregnancy, menstruation, or during sexual act. The scientific community did not accept this link, and it was forgotten until Fleiss' studies in 1897 [3]. Fleiss, a Sigmund Freud's friend, developed a theory of

"reflex nasal neurosis." It was based on a physiological connection between the nose and the genitals. He described distinct 'genital spots' on the inferior nasal concha and suggested medical and surgical treatment for this pathology, but this has all but been forgotten. In 1960, Fabricant published a study on the effects of coitus on nasal temperature. He demonstrated both vasodilation and a rise in the nasal mucosa temperature during sexual intercourse [4].

The nose has defensive and homeostatic functions. The nasal mucosa

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contains sensory, parasympathetic, and sympathetic nerves. Several inputs induce reflex actions in the nose. Inputs such as exercise, pressure on the armpit, cold or warm stimuli on the skin, sexual intercourse, or imagination, can cause the release of neurotransmitters that enhance the secretory and vasomotor activity of the nasal mucosa [5–7]. Sympathetic stimulation brings on noradrenaline release that causes vasoconstriction in the nasal erectile tissue via alpha-adrenergic receptors. Parasympathetic stimulation releases acetylcholine, which leads to vasodilation, glandular discharge, and watery nasal secretions via cholinergic receptors (M3) [8].

The response of the nose to sexual stimuli in the form of sneezing or watery local discharge, is underreported, but it is more common than recognized [9–11]. The parasympathetic stimulus might be the mechanism that triggers such paroxysmal sneezing [9]. We present and discuss a case of 48-years-old male who was treated with endoscopic Vidian neurectomy for sneezing and hydorrhinorrhea during sexual intercourse.

The work has been reported in line with the SCARE criteria [12].

2. Presentation of case

A 48-years-old Caucasian healthy male presented with a two-month medical history of left unilateral nasal discharge, nasal obstruction, sneezing, and facial edema sensation; all mainly present in the morning when he awoke. Those symptoms were more prominent during sexual

intercourse, especially when he reached orgasm, which coincided with a bout of sneezing and watery rhinorrhea. No psychiatric morbidity signs were present. The patient did not have family history of allergic rhinitis, and he had not medical history of head trauma or previous nose surgery.

The symptoms were initially treated with oral and topical antihistamines, and with endonasal topical corticosteroids, but the results were negative. For an extensive diagnostic reasoning, a nasal endoscopy, a head CT and an MRI were carried out. The exploratory nasal endoscopy was normal. The CT detected neither cerebrospinal fluid fistula signs nor skull base defects (Fig. 1). The MRI only revealed a retention cyst in the left maxillary sinus. Laboratory results: the quantification of *Beta*-trace protein (β TP) in the nasal discharge and all allergy tests were negative.

With the clinical suspicion of a pathological imbalance of the autonomic nervous system during sexual arousal mediated by the Vidian nerve (“honeymoon rhinitis”), neurectomy and cautery of this nerve was considered and accepted by the patient. The mucosa of the left middle nasal meatus was subperiosteally dissected by endoscopy. The intervention was done by a surgeon with dilated experience in nasal endoscopy. The sphenopalatine artery was dissected and cauterized at the sphenopalatine foramen. The Vidian nerve and artery were located after removing the sphenoidal process of the palatine bone (Fig. 2). A 2 mm segment of the exposed Vidian nerve was excised, and the remaining nerve was cauterized to achieve a complete sympathetic-parasympathetic denervation of the nasal mucosa (Fig. 3).

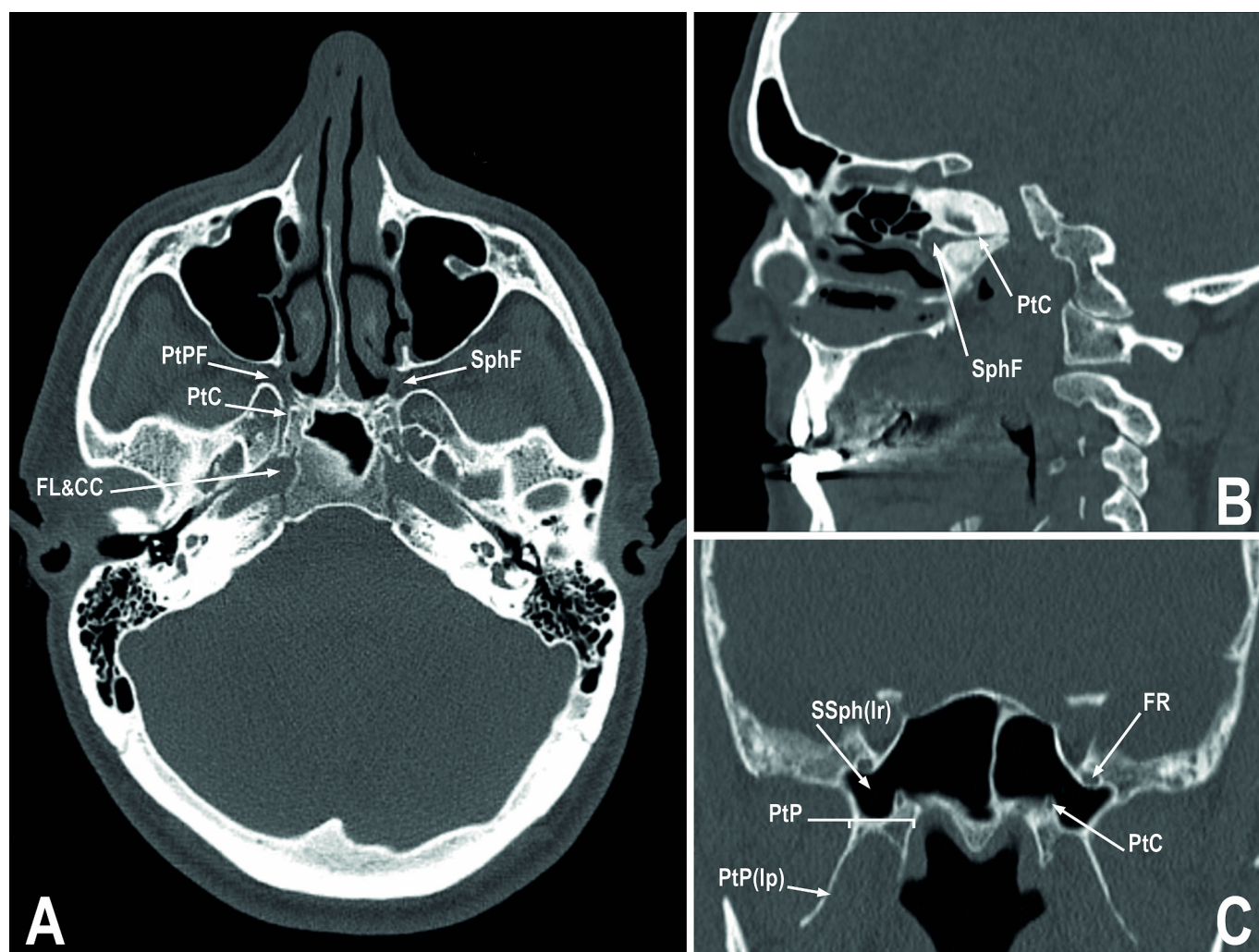


Fig. 1. Images that correspond to the pterygoid canal (PtC) in the CT scan. A (axial view); B (sagittal view); C (coronal view). FL&CC: foramen lacerum & carotid canal; FR: foramen rotundum; PtP: pterygoid process; PtP(Ip): pterygoid process (lateral plate); PtPF: pterygopalatine fossa; SphF: sphenopalatine foramen; SSph(Ir): sinus of sphenoid bone (lateral recessus).

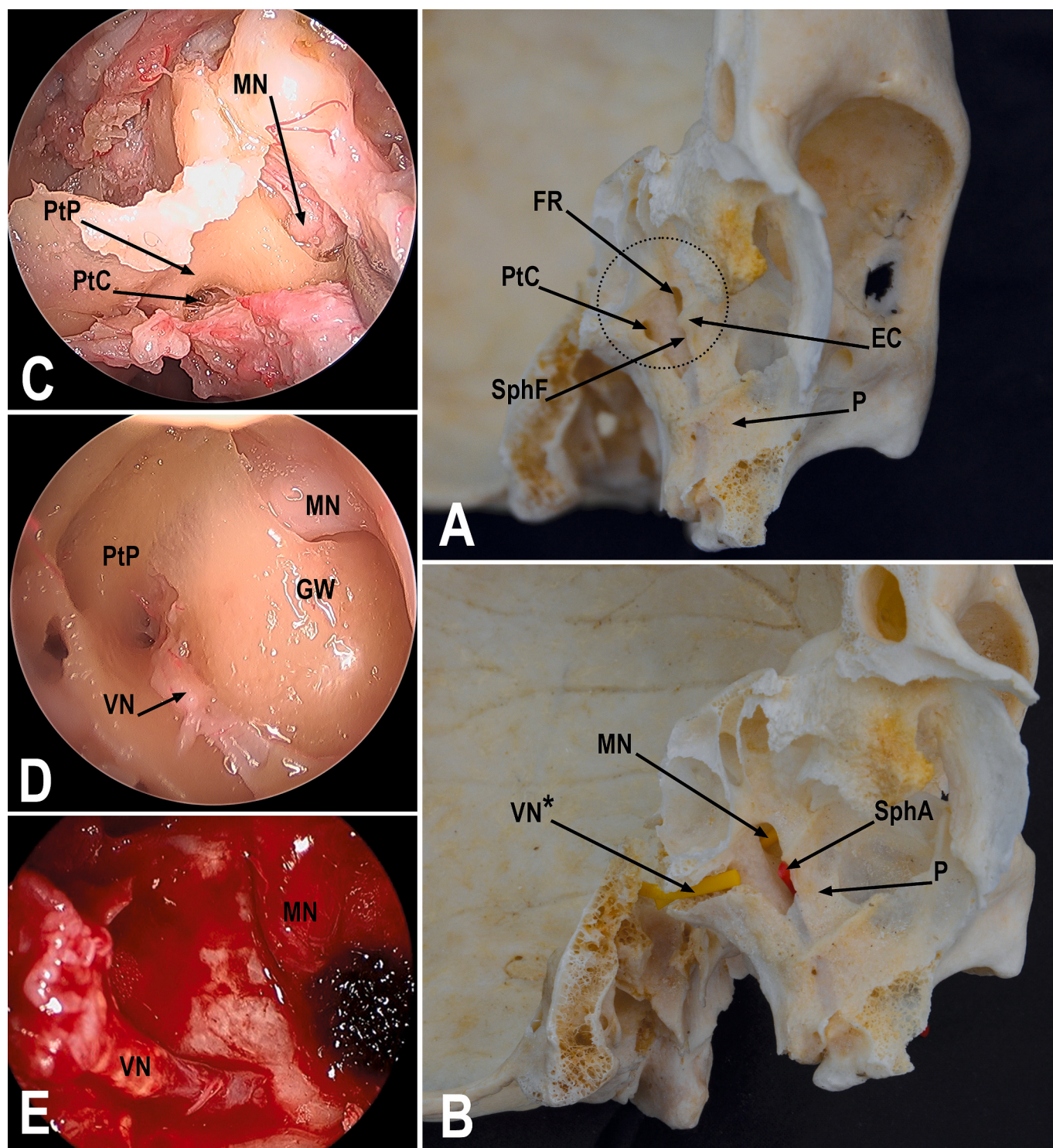


Fig. 2. Images showing the pterygoid canal (PtC) and its relationships at the pterygopalatine fossa. A, B: by a sagittal section of the cranium; C, D: by endoscopy in a fresh-frozen cadaveric specimen; E: by endoscopic surgery in a patient alive. EC: ethmoidal crest of palatine bone; FR: foramen rotundum; GW: greater wing of sphenoid bone; MN: maxillary nerve; P: palatine bone (perpendicular plate); PtP: pterygoid process; SphA: sphenopalatine artery; SphF: sphenopalatine foramen; VN: Vidian nerve; VN*: Vidian nerve exposed after milling the pterygoid canal.

At a follow-up visit at 15 days, the nasal obstruction, sneezing, and rhinorrhea had decreased without showing symptoms either in the morning or during sexual intercourse. No postoperative complications, such as dry eyes or nasal dryness were reported. At the two-year follow-up, the symptoms were still resolved.

3. Discussion

The nerve of the pterygoid canal (Vidian nerve) is formed where the greater petrosal nerve joins the deep petrosal nerve in the foramen lacerum. It runs forward through the pterygoid canal and reaches the pterygopalatine ganglion (Fig. 4).

The greater petrosal nerve is given off from the geniculate ganglion of

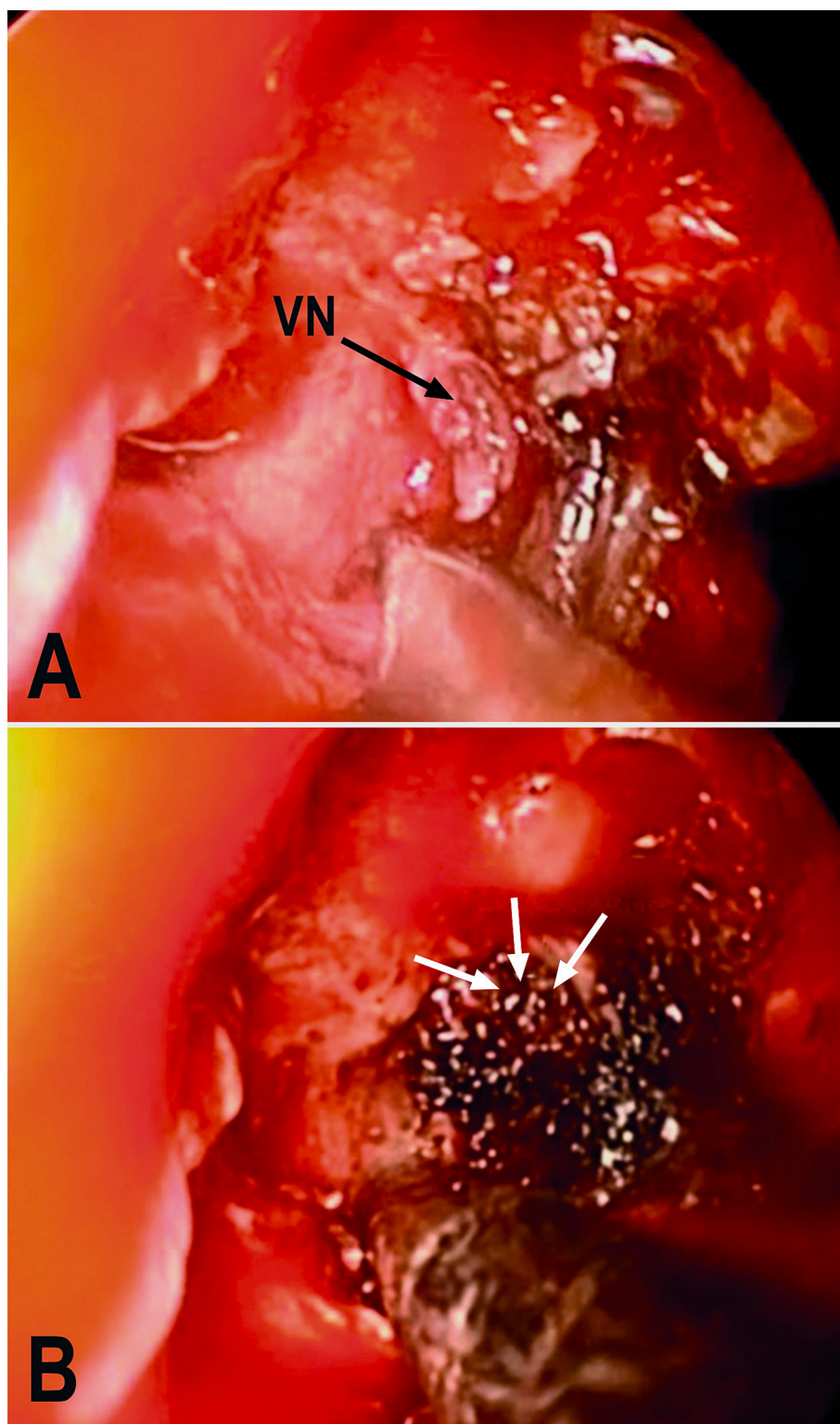


Fig. 3. Images that correspond to the neurectomy of the Vidian nerve (VN) at the pterygopalatine fossa. Arrows show the cauterized Vidian nerve.

the facial nerve, being a branch of the nervus intermedius (nerve of Wrisberg). It carries the preganglionic parasympathetic fibers from the superior salivatory nucleus. Postganglionic axons include secretory fibers for the lacrimal, nasal, and palatine glands. The greater petrosal nerve also contains afferent fibers. Their distribution and function in the nasal mucosa are uncertain [13].

The *deep petrosal nerve* carries sympathetic fibers to the nose. Preganglionic fibers arise from the thoracolumbar spinal cord. They ascend

through the sympathetic trunk and synapse in the superior cervical ganglion. The postganglionic fibers join the carotid plexus. The deep petrosal nerve is given off from this carotid plexus and in the cartilaginous tissue of the foramen lacerum it joins the greater petrosal nerve, thus forming the Vidian nerve. The sympathetic fibers enter the pterygopalatine ganglion without synapsing and provide sympathetic fibers to the branches of the maxillary nerve (V2) [13]. The action of these fibers on the nasal mucosa causes vasoconstriction, thus increasing air

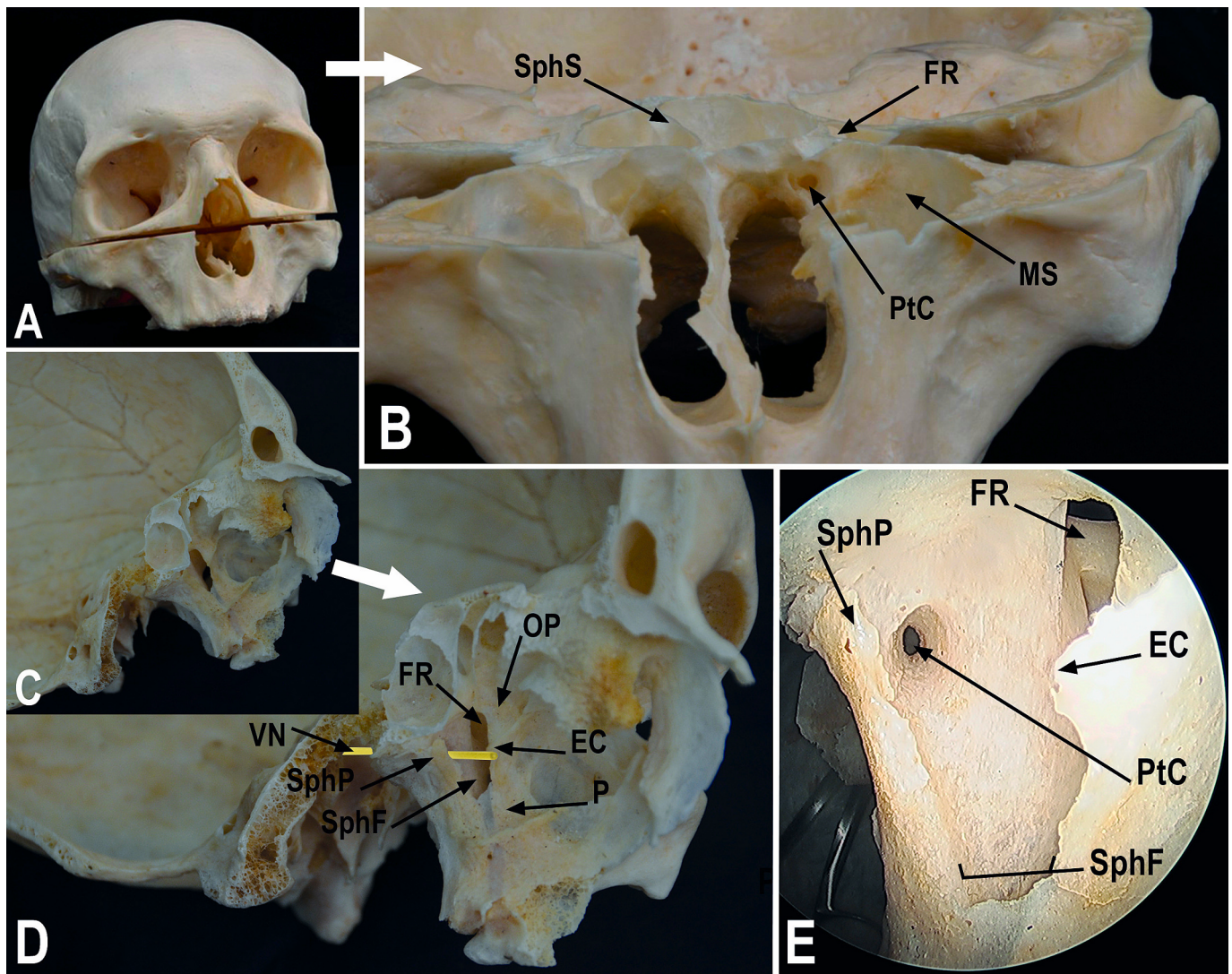


Fig. 4. Axial (A, B) and sagittal (C, D) section images of the cranium to expose the location of the pterygoid canal (PtC) with the representation of the Vidian nerve (VN). E: endoscopic vision of the pterygopalatine fossa through the left nasal cavity. EC: ethmoidal crest of palatine bone; FR: foramen rotundum; MS: sinus of maxilla; OP: orbital process of palatine bone; P: palatine bone (perpendicular plate); SphF: sphenopalatine foramen; SphP: sphenoidal process of palatine bone; SphS: sinus of sphenoid bone.

flow. The postganglionic sympathetic and parasympathetic fibers also provide innervation to the lacrimal gland through the zygomatic nerve [13]. The nasal cycle is the alternation of congestion and decongestion with changes in the airflow [14]. Cycling of the resistance airflow is reported in 80 % of subjects, and it seems to depend on sympathetic innervation because the nasal cycle is abolished when the cervicothoracic ganglion is blocked. However, the disruption of parasympathetic fibers also affects it. This fact demonstrates that the nasal cycle needs the autonomic nervous system to be balanced [14,15].

Sexual intercourse and orgasm also induce a response in the nasal mucosa associated with autonomic stimulation and increased parasympathetic activity, mainly during sexual arousal and the culmination of orgasm. In contrast, there is a sympathetic preponderance after orgasm. However, sneezing in response to either sexual imagination or following orgasm is an underreported phenomenon. A physiological mechanism of parasympathetic summation seems to cause this unusual response [10]. This imbalance with parasympathetic overactivity can occur in some patients by releasing the mast cell mediators that provoke postcoital rhinitis [11].

Sexual intercourse is a poorly recognized trigger for postcoital asthma, rhinitis and sneezing [1,4,9–11]. During sexual intercourse, in

these patients, the parasympathetic outflow has similar effects on the penis, clitoris, and nasal mucosa, with venous dilation of the corpus cavernosum and nasal secretion. The result of this parasympathetic-sympathetic imbalance during sexual intercourse is known as “honeymoon rhinitis.”

The parasympathetic part of the autonomic nervous system is not arranged segmentally. The vagal region in the neural tube is near the putative future sneeze center. The vagal center is the hub of all parasympathetic innervation. There is evidence of persisting links between different components of the parasympathetic nervous system, whereby stimulation of one parasympathetic response will lead to other parasympathetic responses (parasympathetic summation phenomenon). Some examples are the parasympathetically mediated pupillary constriction, which can be accompanied by secretion from the nasal mucosa and sneezing in response to bright light, or bradycardia and hypotension during sleep that can be accompanied by penile erection. In parasympathetic summation, any trigger that stimulates a parasympathetic response also makes references to their embryonic origin in the vagal nuclei, which can, in turn, stimulate other parasympathetic responses [10]. During sexual stimulation, there is a parasympathetic activity for venous dilation in the corpus cavernosum. This

parasympathetic outflow can include an efferent response in the greater petrosal nerve, which can trigger sneezing and mucus production in the nose through the Vidian nerve [16].

Goldin-Wood [17,18] proposed the Vidian neurectomy by a trans-antral approach to treat vasomotor rhinitis, polyposis, and sinusitis. The reported results have been heterogeneous, and relapses of symptomatology are frequent. Before the use of endoscopy, locating the Vidian nerve and the pterygoid canal was a challenge. Thus, neurectomies were often not complete and the technique was almost completely abandoned. The work of Kamel and Zaher [19] demonstrating the endoscopic Vidian neurectomy in cadaver and the use of preoperative CT guidance led us to a more precise surgical technique with better results and low morbidity [20].

The surgical procedure for treating refractory vasomotor rhinitis has poor long-term outcomes because of several challenges in locating the Vidian nerve. It has also been proposed as a treatment for chronic cluster headaches, chronic epiphora, senile nasal drip, and crocodile tears [21], but in the literature reviewed, we have not found any reference to the Vidian neurectomy as a treatment for “honeymoon rhinitis”, probably due to it is an underreported condition because of it is embarrassing for patients. Complications such as dry eye (35 %–72 %), paresthesia in the V2 dermatome (8.2 %), palatal or gingival numbness (6.72 %), and nasal crusting (3.7 %) have been reported. A meta-analysis of 32 studies that included 2185 patients concluded that endoscopic Vidian neurectomy remains feasible for medically recalcitrant rhinitis [21,22]. In this way, the introduction of endoscopic techniques has meant a clear improvement in the outcomes [23,24].

4. Conclusion

“Honeymoon rhinitis” is an underreported symptom. The etiology seems to be due to an imbalance in the autonomic nervous system. We suggest that Vidian neurectomy could be indicated when other treatments fail. The reported case was unilateral, and we have not found any other similar cases in the reviewed literature. The good response after Vidian neurectomy supports the hypothesis that a parasympathetic-sympathetic imbalance was the cause of this disorder. The main limitation of this study is that it is based on a single case.

Author contribution

Humbert Massegur: conceptualization, methodology, formal analysis, investigation, writing original draft, supervision.

Ana Carrera: conceptualization, resources, data curation.

Juan Ramon Gras-Cabrerizo: data curation, formal analysis, supervision.

Richard S. Tubbs: conceptualization, writing review and editing.

Francisco Reina: conceptualization, formal analysis, writing review and editing, supervision.

Consent

Written informed consent was obtained from the patient for publication and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

The authors declare the study of a single case report is exempt from ethical approval of the health institution (Sant Pau Hospital Research Ethic Committee CEI). The authors state that every effort was made to follow all local and national laws that pertain to the use of case reports in research.

Guarantor

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Conflict of interest statement

The authors declare we have no conflicts of interest.

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