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Sphenopalatine ganglion deficit syndrome: An unusual complication after septoplasty

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ABSTRACT

Background: In 1908 Sluder described a clinical picture of unilateral facial pain, lachrymation, rhinorrhea and mucosal congestion deriving from the Sphenopalatine Ganglion (SPG) irritation. We described a case of monolateral xerophthalmia, dry palate and mouth, deriving from SPG lesion after septoplasty that we called "SPG Deficit Syndrome".
Methods: In our study a woman complaining functional nasal disorders and Computer Tomography (CT) images of a huge condro-vomerian septal spur in right nasal cavity, was underwent to septoplasty. After surgery she complained monolateral xerophthalmia, xerostomia and migraine.
Results: We formulated hypothesis of parasympathetic postgangliar nerve transmission interruption due to a lesion of effector fibers, supported by post-operative CT images of posterior wall of maxillary sinus lesion and by endoscopic evaluation of dryness of palatal mucosa and right nasal cavity.
Conclusion: To the best of our knowledge this is the first case of this kind of symptomatology reported as complication after septoplasty.

1. Introduction

In 1908, Sluder proposed that a high-grade inflammatory reaction in the posterior ethmoid and sphenoid sinuses may be involved in certain cases of unilateral facial pain associated with lachrymation, rhinorrhea, and mucosal congestion. These symptoms are related to the inflammation or neuropeptides release that cause vessel dilation and/or activation of the trigeminal nociceptor fibres into the sphenopalatine ganglion (SPG) [1]. The SPG, even called Meckel's ganglion, is the largest extra cranial neural structure located into pterygopalatine fossa (PPF). The ganglion has sensory, motor and autonomic components, for the sensibility of the palate, the motility of the velum palatinum elevator muscle, but the most representative fibers are the parasympathetic fibers, which inputs derive from the superior salivatory nucleus (SSN) in the brainstem. The efferent postganglionic fibers provide secretomotor function to the mucous membrane of the nose, soft palate, tonsils, uvula, roof of the mouth, upper lip and gums, upper part of the pharynx, lacrimal

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gland, and meningeal vessels [2] (Fig. 1a).

The aim of our study is to describe the physiopathological bases underlying an unusual syndromic clinical picture characterized by monolateral xerophthalmia, dry palate and mouth, deriving from the lesion of the SPG after septoplasty. We have called this lesional picture as "SPG Deficit Syndrome" to underline the differences with the one described by Sluder due to an irritative stimulus. To the best of our knowledge this is the first time description of such a kind of syndromic clinical picture reported as a complication after septoplasty.

2. Case report

A 43 years-old woman, who complained functional nasal disorders with a sense of obstruction and breathing difficulty, was admitted to our Maxillo-Facial Surgery Unit on July 2013 for elective septoplasty. To the medical history no prior trauma or surgery was declared, she only suffered for Gastro-Esophageal Reflux Diseases (GERD), under treatment with Proton-pump inhibitors (PPIs). Pre-surgical Computer Tomography (CT) showed a huge condro-vomerian septal spur in the right nasal cavity (Fig. 1b and c).

On September 2013, a closed septoplasty was performed under general anesthesia. Through a *trans*-columellar incision, the right septal spur was removed with use of a nasal osteotome. Total operation time was 65 min, and no anomalies were found during the surgical procedure. Nasal packing was applied, and the awakening from anesthesia occurred without complications. After nasal packing removal, at day three, the patient began to complain a symptomatology characterized by right xerophthalmia, dryness and hypoesthesia of palate and nose. The patient also began to suffer of frequent episodes of right migraine without aura, never complained before. Post-surgical CT showed a linear bone lesion at the posterior wall of the maxillary sinus extended to the PPF. The PPF impairment, appearing blocked due to scattered bone fragments, was evident (Fig. 2a–c).

She also practiced a Schirmer test with positive result for hypo-lacrimation (<5mm), therefore she was admitted to regular clinical follow-up evaluation. The referred symptomatology was supported by the endoscopic examination performed after a medium turbinectomy to improve PPF exposure and attempt removal of incarcerated fragments. On videoendoscopy, the mucous membranes in the right nasal cavity appeared dry and covered with abundant crusts adhering to the surface. (Fig. 2d). At the anterior wall of the pterigopalatine fossa abundant scar tissue was appreciated by subverting the normal anatomy. Frequent clinical checks were necessary for crusts removal, despite the prescription of nasal washings with saline solution and hyaluronic acid spray based. Due to the persistence of symptoms and non-responsiveness to migraine therapy, the patient was candidate for corticosteroid (CCS) infiltration in the right PPF via endonasal endoscopic approach under local anesthesia. After CCS infiltration the patient showed an improvement in migraine, while a persistence of xerophthalmia and nasal and palatal dryness was assessed. At 22 months follow-up after second surgery, the syndromic picture remained unchanged. The patient is currently under ophthalmological and maxillofacial follow-up for nasal crusts removal and prevention of keratitis. The study was conducted according to Helsinky's declaration.

3. Discussion

The SPG is an autonomic ganglion, communicating with the first and second division of the trigeminal nerve, the facial nerve, and the carotid plexus. It has also been labeled Meckel's ganglion, the pterygopalatine ganglion (PPG), and the nasal ganglion, but the SPG may be the preferred historical term and has now long been entrenched in the medical literature. It is allocated in the pterygopalatine fossa; this is a small pyramidal space, upside down, 2 cm high and 1cm wide, situated behind the posterior wall of the maxillary sinus,

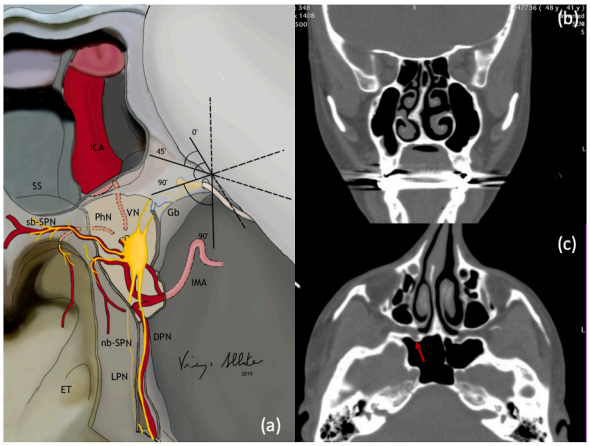

Fig. 1. a) Anatomical illustration of the SPG. VN Vidian Nerve; PhN Palatovaginal Nerve; Gb ganglionic branches; DPN and LPN Discending palatine nerve and Lasser palatine nerve; sb-SPN septal branches of the sphenopalatine nerve; SS sphenoid sinus; ICA Internal Carotid Artery; ET Eustachian tube; b) Preoperative coronal CT scan showing the right septal chondrovomerian spur; c) Preoperative axial CT scan showing the integrity of the right pterygopalatine fossa (red arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

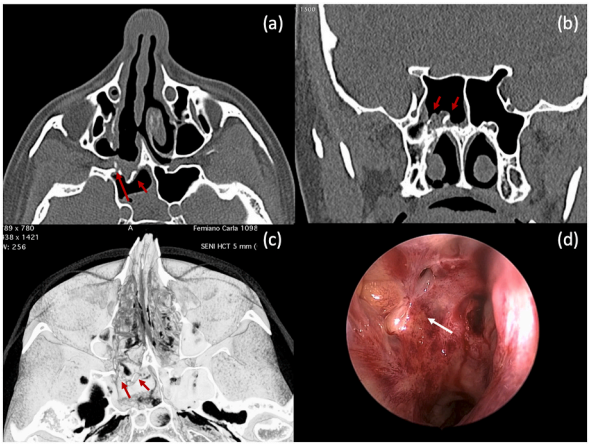

Fig. 2. Postoperative CT scans showing the collapse of the right pterygopalatine fossa with scattered bone fragments (red arrows); a) CT axial view; b) CT coronal view; c) 3D axial reconstruction; d) Postoperative endonasal endoscopic vision of the right nostril. The picture shows in the right pterygopalatine fossa the subversion of the normal anatomy and abundant scar tissue that covers the sphenopalatine foramen (white arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

anterior to the medial plate of the pterygoid process, and lateral to the perpendicular plate of the palatine bone; superiorly, the pterygopalatine fossa is limited by the sphenoid and laterally it communicates with the infratemporal fossa. Superiorly, the fossa communicates with the orbital apex through which some nerve branches reach the lacrimal gland. The posterior wall of the fossa has three important openings: superolaterally the foramen rotundum at 0° degree, which transmits the second branch of the trigeminal nerve; infero-medially the Vidian (pterygoid) canal at 45°, related to the anonimus nerve (Deep and Greater petrosal nerves) and the pterigovaginal canal [aka pharyngeal canal] at 90°(Fig. 1a).

The SPG is located posterior to the middle turbinate and is few millimeters deep to the lateral nasal mucosa.

The ganglion has sensory, motor and autonomic components, but the most representative fibers are parasympathetic, which inputs derive from the superior salivatory nucleus (SSN) in the brainstem. The preganglionic parasympathetic fibers pass through the intermediate nerve and reach the PPF by the greater superficial petrosal nerve (GSPN) across the Vidian channel. The GSPN also carries the gustatory sensitivity that reaches the palate through the palatine major in inferior channels. The parasympathetic secretomotor fibers synapse in the SPG and the postsynaptic fibers distribute to the mucous membrane of the nose, soft palate, tonsils, uvula, roof of the mouth, upper lip and gums, upper part of the pharynx, lacrimal gland, and meningeal vessels [2].

For its innervation, both sympathetic and parasympathetic, the SPG is believed to play a part in the headache pain and cranial autonomic symptoms associated with cluster headache, which is a result of activation of the trigeminal-autonomic reflex. In cluster headache, post-ganglionic parasympathetic fibres from the SPG that innervate the cerebral and meningeal blood vessels are activated releasing neuropeptides that cause vessel dilation and/or activation of the trigeminal nociceptor fibres in the meninges, which is perceived as referred pain from the head by the sensory cortex [3].

In 1908 Sluder was the first to highlight the role of the SPG in neuralgic syndromes of the face. Sluder reported that patients who had refused surgery for an active ethmoido-sphenoidal inflammation, developed sphenopalatine ganglion neuralgia (SPGN) later [4].

Thus, Sphenopalatine ganglion neuralgia or Sluder's neuralgia (SPGN) is a type of facial neuralgia, defined as a complex symptom consisting of neuralgic, motor, sensory, and gustatory manifestations. SPGN refers to intermittent episodes of vasomotor hyperactivity causing conjunctival injection, lacrimation, serous nasal discharge sensory disturbances of the palate and oropharynx with distorted gustatory sensations.

Sluder describes a symptomatology perfectly mirrored to that reported by our patient. The mechanism on the base of this manifestations is certainly different by Sluder's described mechanisms, so we have supposed that there was an interruption of the parasympathetic postgangliar nerve transmission due to a lesion of the effector fibers. This hypothesis is supported both by post-operative imaging that showed a damage on the right PPF, and by endoscopic direct evaluation of dryness of nasal and palatal mucous membranes (Fig. 2a–d). Given the characteristics that identify this syndrome, we could call it "SPG deficit syndrome", to underline the opposition between the lesional mechanism and the irritating mechanism described by Sluder.

From the pathophysiological point of view our hypothesis is reflected in some syndromes described as complications after SPG blockade for the treatment of complex neuralgic syndromes of the face.

Narouze et al. listed a set of SPG post-ablation radiofrequency side effects, for the treatment of cluster headaches, that seem to be compared with our case [5].

4. Conclusion

We can conclude that even septoplasty is not free of consequences. "SPG deficit syndrome" can be an extraordinary and unusual complication to observe.

To the best of our knowledge, this is the first case report of this symptomatology as a complication after septoplasty. The main drowback of our study refers the absence of MRI due to the patient's refusal and the lack of treatment option for this syndrome, that may be the subject of other our future studies.

Patient consent

The patient has consented to the submission of the case report.

Ethical approval

No ethical approval was needed. Signed patient consent was all that was required by the hospital.

Declaration of competing interest

The authors declare that they have no conflict of interest.

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