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Navid Ahmady-Roozbahany
Shahid Beheshti University of Medical Sciences

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Nasal and paranasal sinus anatomical variations in patients with rhinogenic contact point headache

Navid Ahmady Roozbahany [a,*], Semiramis Nasri [b]

[a] Otolaryngology-Head and Neck Surgery, Urmia Medical University, Urmia, Iran
[b] Private Practice, Urmia, Iran

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ABSTRACT

Objective: To define anatomical variations that may lead to rhinogenic contact point headache.
Methods: Paranasal sinuses CT scan and medical records of sixty-five patients who underwent a successful endoscopic surgery for rhinogenic contact point headache reviewed.
Results: Eleven distinct anatomical variations were found in patients with rhinogenic contact point headache. All of them were surgically curable.
Conclusion: There are multiple anatomical situations that may lead to rhinogenic contact point headache and each one has its own characteristics. Treatment plan should be personalized for every patient considering the diagnosed anatomical variation.

1. Introduction

Intranasal contact points refer to a contact between two opposing intranasal mucosal surfaces. These contacts occur between nasal septum and lateral wall of the nose. Intranasal contact points are reported to be present in about 4% of noses [1,2]. It is now completely accepted that these contact points can be a cause of headache and facial pain. The relation between nasal structures and headache was first described in 1948, when Wolff in his book stated that nose and paranasal structure can be sources of headache [1]. Stammberger and Wolf described the role of substance P in pathophysiology of rhinogenic contact point headache [2]. Their work also demonstrated that this kind of headache in not solely introduced by an abnormal middle turbinate but every intranasal mucosal contact may cause a referral pain. The selection of patients who might benefit from a surgical procedure is critical. The reported success rate from surgery is only 30–60% [3]. Although many anatomical variations with a contact between nasal septum and nasal lateral wall can be assumed, the distinct anatomy of nose and paranasal sinuses allows several common conditions to occur; anyway these anatomical variations have not been systematically studied yet.

Abbreviations: RCPH, rhinogenic contact point headache; PNS, paranasal sinuses; VAS, Visual Analogue Score; MTCB, middle turbinate concha bullosa; STCB, Superior Turbinate Concha Bullosa; SP, substance P.

* Corresponding author at: Otolaryngology Department, Imam Hospital, Ershad Street, Urmia, Iran. Tel.: +98 912 3185283; fax: +98 441 2367983.
E-mail address: navidar@yahoo.com (N.A. Roozbahany).

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2. Materials and methods

A retrospective review of paranasal CT scan image of patients who underwent endoscopic nasal surgery for rhinogenic contact point headache (RCPH) was conducted. They were candidate of surgery because of a clinical history, physical examination and imaging in favor of the diagnosis of RCPH. All patients had headache or facial pain that was justifiable with intranasal mucosal contact. In pre-operative CT scans of all patients, a distinct contact between septum and one of lateral nasal wall elements could be found. All these patients had positive shrinkage test which is relief of pain and discomfort after prescription of an anesthetic and vasoconstrictive solution on the intranasal contact area and finally, all patients had significant relief of symptoms after surgical removal of mucosal contact points.

In a two years period from November 2009 until December 2011, we operated 123 patients with the diagnosis of RCPH. Our exclusion criteria for this study were any extent of sinonasal inflammatory disease that was present in 32 patients, incomplete relief of symptoms (11 patients) and post-operative follow-up less than 6 months (15 cases).

Sixty-five patients enrolled the study. The study had a retrospective nature. It was designed after treatment and follow-up of all patients; for this reason, requirement of a written informed consent waved. Anyway, all patients had signed consent to use of clinical information at the time of admission and it was present in the medical record of the ones who enrolled this study.

The paranasal sinuses (PNS) CT scans were performed in coronal plans with 5 mm intervals and looked carefully for

Table 1
Anatomical variations in patients with rhinogenic contact point headache and characteristics of headache preoperatively (VAS: Visual Analogue Score).

Anatomical variation	Number	Percent	Mean severity (VAS)	Mean duration (hours/24 h)	Mean frequency (days/month)
Septal deviation	12	18.50%	6.7 ± 0.87	6.0 ± 2.7	6.1 ± 4.9
Septal spur	7	10.80%	7.7 ± 1.2	12.3 ± 6.1	17.0 ± 6.8
Septum bullosa	2	3.10%	7.0 ± 0.0	4.5 ± 0.7	6.0 ± 5.7
Medialized middle turbinate	2	3.10%	6.5 ± 0.7	2.5 ± 0.7	5.5 ± 0.7
Paradoxical curvature of middle turbinate	3	4.60%	7.0 ± 1.0	5.0 ± 2.6	4.3 ± 1.5
Concha bullosa of middle turbinate	19	29.20%	7.1 ± 1.0	8.2 ± 5.7	8.9 ± 5.1
Lamella bullosa	2	3.10%	8.5 ± 0.7	17.5 ± 3.5	12.5 ± 3.5
Medialized superior turbinate	3	4.60%	6.7 ± 0.6	6.7 ± 1.5	5.0 ± 1.7
Concha bullosa of superior turbinate	6	9.20%	7.8 ± 0.7	9.3 ± 3.5	12.6 ± 6.9
Large bulla ethmoidalis	7	10.80%	7.4 ± 1.3	5.0 ± 2.6	5.1 ± 1.9
Hyperaerated posterior ethmoidal cells	2	3.10%	6.0 ± 1.4	4.5 ± 0.7	6.0 ± 1.4
Total	65	100.00%	7.1 ± 1.0	7.6 ± 5.0	8.6 ± 6.0

anatomical variations and intranasal contact points. The clinical history of patients including a questionnaire about the characteristics of pain and accompanying symptoms and also results of preoperative studies were collected from records of patients. The severity of pain had been assessed using a Visual Analogue Score (VAS) in which 0 was no pain and 10 was the worst imaginable pain.

We performed Chi Square test using SPSS ver. 19.0 when a statistical analysis was needed.

3. Results

Sixty-five patients (30 male/35 females – mean age 34 ± 3.4 years) studied. All of them had headache or facial pain. The unset time of the pain was ranged from 6 months to 17 years (mean 5 ± 2.3 years) before treatment. The pain of 21 patients (32.3%) was continuous and in the remaining cases, the frequency of pain was ranged from twice a month to three times a week (Table 1). 14 patients (21.5%) had a pulsating pain, 19 patients (29.2%) had photophobia and 14 ones (21.5%) had phonophobia. 21 patients (32.3%) had received unsuccessful medical treatment for migraine. The average headache severity in our patient fell from 7.1 to 1.2 and only five patients still had degrees of pain after surgery. In these five cases, severity, duration and frequency of pain were decreased significantly and they were satisfied by the results of the treatment.

In 12 patients (18.5%), septal deviation was the only cause of mucosal contact. In 5 persons, septal deviation was secondary to a trauma and septal fracture (Fig. 1). In these patients septal deviation was severe or high enough to make a contact between middle or superior turbinate. Most of patients in this group had pain in forehead in the ipsilateral side.

7 patients (12.1%) had a septal spur that was in close contact with lateral wall of the nose (Fig. 2). These patients had pain in forehead and orbital region. They had more severe pain in comparison with the ones who had a simple septal deviation (Table 1) but this difference was not statistically significant. On the other hand, frequency and duration of pain in these patients was significantly higher ($p < 0.005$).

Septum bullosa (Fig. 3) was found in 2 patients (3.1%). We considered septum bullosa as the cause of RCPH when a lamella of septal air cell was the only anatomical element which had close contact with lateral wall. The characteristics of pain were very similar to septal deviation but there were some differences in diagnosis and treatment.

The RCPH in two patients (3.1%) was because of a contact between a medialized middle turbinate and the nasal septum (Fig. 4). This anatomical variation causes the less severe and durable pain among our patients. In 3 patients (4.6%), paradoxical curvature of middle turbinate seemed to cause the mucosal contact (Fig. 5). The most common anatomical abnormality among our

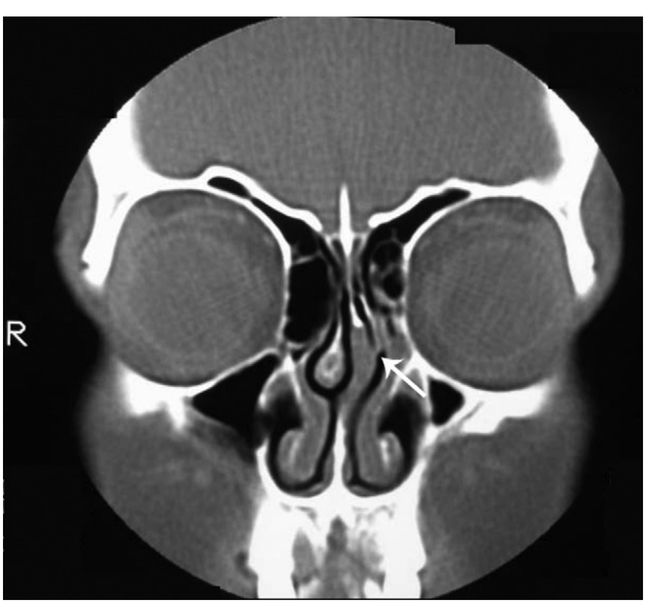

Fig. 1. Septal deviation. Contact between septum and left middle turbinate (arrow).

Fig. 2. Septal spur. Contact between the spur and right middle turbinate (arrow).

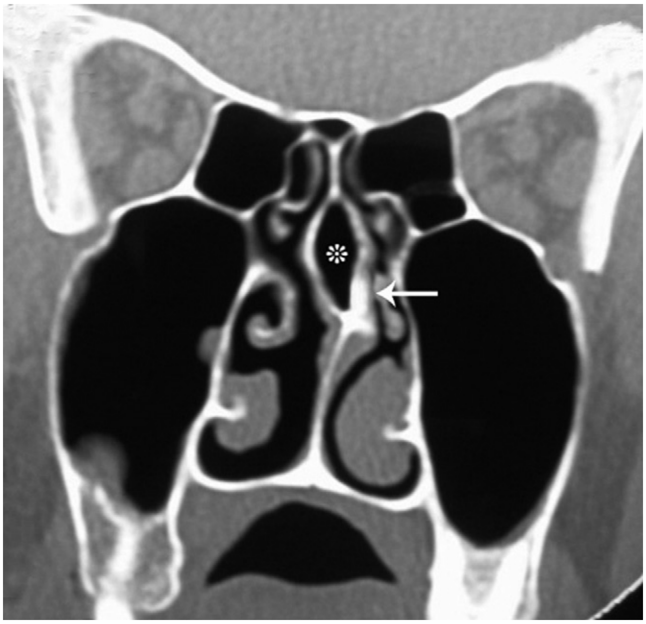

Fig. 3. Septum bullosa (star). Contact between left lamella of septum bullosa and middle turbinate (arrow).

patients was middle turbinate concha bullosa (MTCB) which was found in 19 cases (29.2%). Concha bullosa of middle turbinate could be small or huge, unilateral or bilateral and uni-chambered or multi-chambered (Fig. 6). Lamella bullosa which is pneumatization of vertical lamella of middle turbinate was present in 2 patients (3.1%) (Fig. 7). These patients had the most severe, frequent and durable pain among our patients (Table 1). The pain was felt in medial cantus and forehead in both cases.

The superior turbinate was abnormally medialized (Fig. 8) and was in contact with upper septum in 3 cases (4.6%). In 6 patients (9.2%) we found concha bullosa of superior turbinate (STCB) in one or both sides (Fig. 9). The frequency of pain in patients with STCB was significantly more than the ones with medialized superior turbinate ($p < 0.005$). It was also more durable and more severe but the later differences were not statistically significant.

Fig. 4. Bilateral medialized middle turbinates. Contact between middle turbinates and septum in both sides (arrows).

Fig. 5. Paradoxical curvature of left middle turbinate (arrow).

Seven patients (10.8%) had a large bulla ethmoidalis. In two cases, the bulla was large enough to extrude even outside of middle meatus (Fig. 10) and make a contact with the septum by itself. In the remaining cases, the bulla was pushing the vertical lamella of middle turbinate and was placing it in contact with septum. The characteristics of pain in these patients were very similar to the ones with a paradoxical middle turbinate.

In 2 patients (3.1%), posterior ethmoidal cells (Fig. 11) were hyperaerated and there was a contact between medial wall of these cells and septum. The characteristics of pain in these patients were comparable to with the ones who had septal deviation or medialized middle turbinate.

4. Discussion

Rhinogenic contact point headache (RCPH) is a referral pain that arises from contact between nasal septum and lateral nasal wall. Wolff described the concept of referral headaches due to intranasal contact points and proposed that headaches or facial pain could occur secondary to contact between the turbinates and other regions of the nasal cavity [1]. Some authors called this phenomenon middle turbinate headache syndrome [4,5]. It is now accepted that contact point pains are mediated by the stimulation of intranasal polymodal receptors that are innervated by afferent C fibers of ophthalmic and maxillary branches of trigeminal nerve. The pain is projected and felt in the cutaneous distributions that are corresponding dermatomes of these two branches [1]. Substance P (SP) has a known role in pathogenesis of RCPH [2]. SP is an important neuropeptide that can be identified in the nasal mucosa. Release of SP in the central nervous system may cause referred pain, and also some local reflexes are mediated by SP. These reflexes cause vasodilatation, plasma extravasation and hypersecretion. When SP is released at perivascular sites, vasodilatation, plasma extravasation and perivascular inflammation can cause headache similar to migraine without aura [1].

The receptors can be stimulated by chemical and caloric and mechanical irritants. The pressure exerted on nasal mucosa can be enough to trigger an SP-mediated pain sensation via afferent C fibers [1,2,6]. It seems that contact between mucosal surfaces in the nose would elicit more pain than chronic infection or inflammation. Measurement of the concentration of SP in human

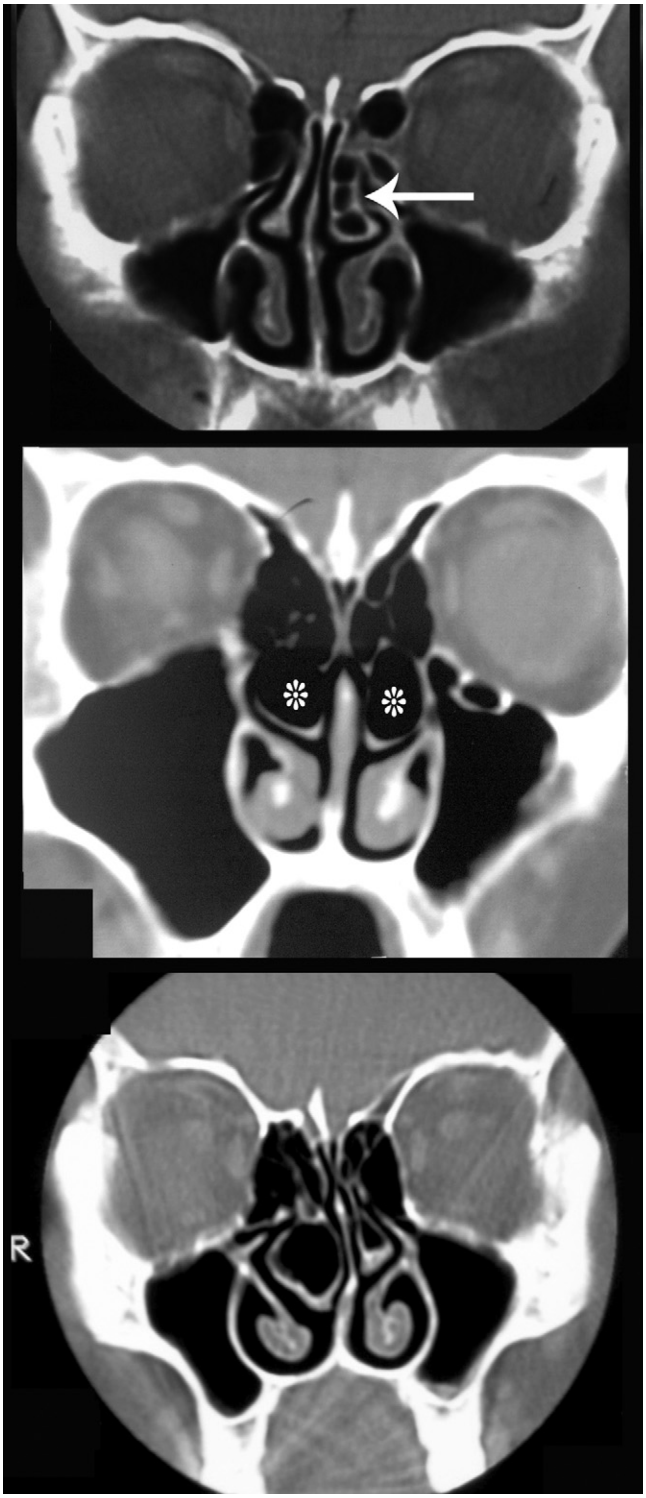

Fig. 6. Concha bullosa of middle turbinate. Top: Unilateral three chambered concha bullosa (arrow). Middle: Bilateral symmetric concha bullosa (stars). Bottom: Bilateral asymmetric concha bullosa.

nasal mucosa has shown that normal mucosa has higher concentrations of SP than chronic hyperplasic mucosa or polyp tissue [2]. This justifies why contact point headaches are almost always seen in cases without rhinosinusitis.

Diagnosis of contact point headache requires a multidisciplinary approach. Patients with headache and without findings of inflammation in the nose and sinuses should be examined to

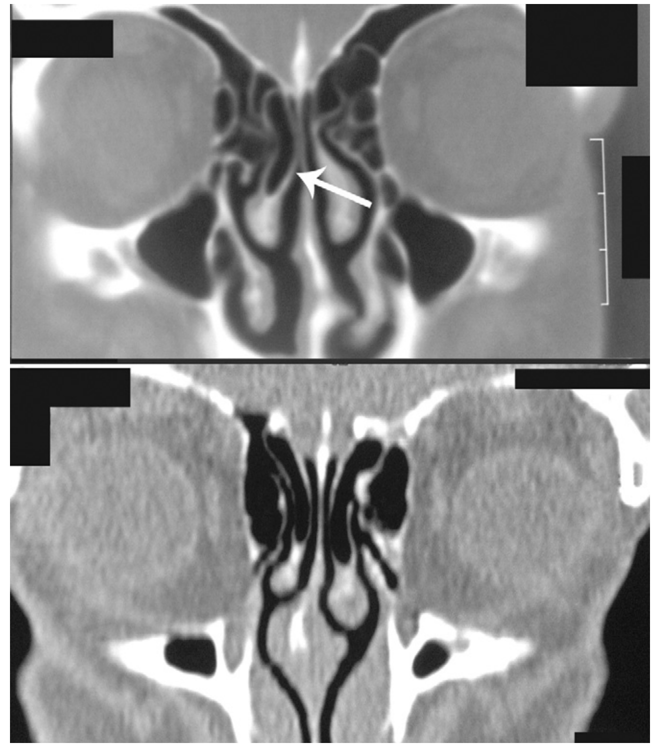

Fig. 7. Lamella bullosa. Top: Unilateral (arrow). Bottom: Bilateral.

Fig. 8. Medialized superior turbinate (arrow). Contact between right superior turbinate and septum.

exclude other causes of headache and evaluation for intranasal contact points should be included. The combination of CT scan with diagnostic nasal endoscopy provides the maximum information [1].

Behin et al. found intranasal contact points other than middle turbinate and speculated that contact point headaches may also be caused by the contact between the septum and the superior turbinate or medial wall of the ethmoidal sinus [7]. Huang et al. and Welge-Luessen et al. demonstrated that following identification of the mucosal contact area, RCPH can be cured with surgical management [3,8].

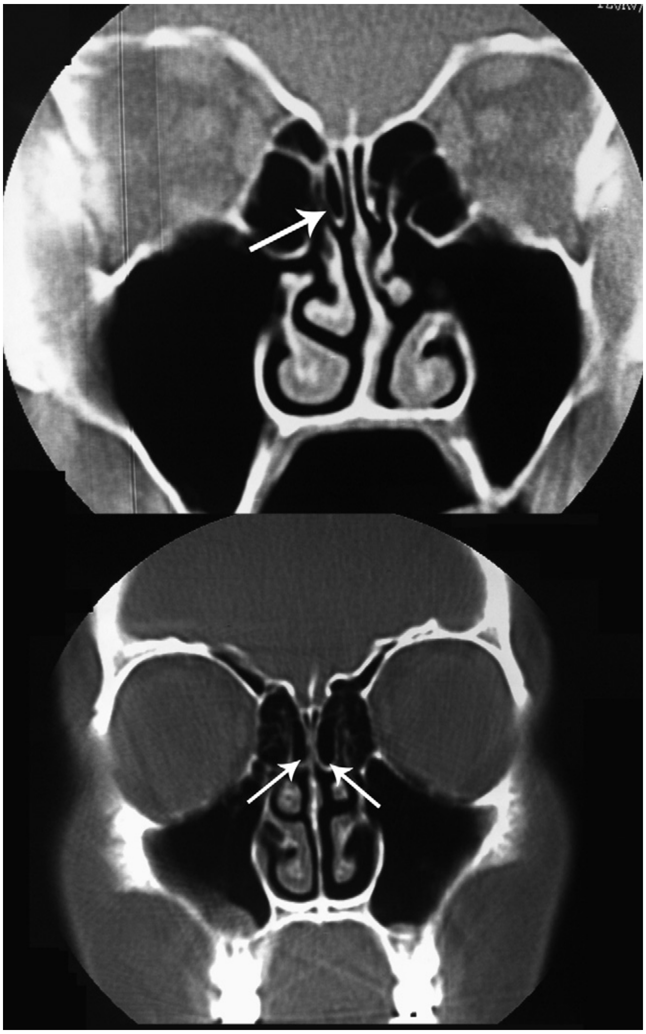

Fig. 9. Superior turbinate concha bullosa. Top: Unilateral (arrow). Bottom: Bilateral (arrows).

Fig. 10. Large bulla ethmoidalis. Both pictures belong to a single patient. Top: Anterior to the middle turbinate. The bulla ethmoidalis (stars) protrudes outside of the middle meatus and is in contact with septum (arrows). Bottom: The relation between the large bulla ethmoidalis and the middle turbinate.

We reviewed 61 CT scan from patients diagnosed as having RCPH. This diagnosis was made considering the medical history, symptomatology and physical examination. These patients were candidates for surgery due to results of endoscopic examination, pre-operative CT scans and positive shrinkage test. The diagnosis confirmed after a successful surgery. The surgery was individualized for each patient and its aim was to remove each and every mucosal contact point that a patient might have. Patients enrolled in this study only when post-operative follow up after at least 6 months revealed significant relief so we assume that this relief is an important indicator for presence of the intranasal mucosal contact point which was the main cause of patients' symptoms. This is why the response to surgical treatment was considered as an inclusion criterion. We can also propose that all abnormalities observed in pre-operative CT scans had clinical importance.

We found septal deviation as the cause of mucosal contact in 12 patients. The convex side of septum was in contact with ethmoidal cells, superior turbinate or middle turbinate. These patients had no other anatomical variations such as concha bullosa and/or medialized turbinates and the contact point was removed only with a conventional septoplasty. We believe that a careful examination of nasal cavity and observation of pre-operative imaging is necessary prior to performing a septoplasty in RCPH patients. All parts of nasal septum especially higher portions must be addressed during surgery to avoid insufficient operation.

Fig. 11. Bilateral hyperaerated posterior ethmoidal cells. Contact between medial wall of posterior ethmoidal cells and septum (arrows).

In 7 cases, we found a septal spur in the site of mucosal contact. We consider septal deviations and septal spurs as separate groups because the characteristic of pain was different between these two groups. All patients with septal spur had pain in medial canthus as well as forehead while periorbital pain was less common among patient with septal deviation. In addition, septal spur cases had more frequent and durable pain than the ones with septal deviation. Operation on these patients needs more care as the mucoperichondrium at the site of the spur is prone to tearing and a septal perforation may happen.

In two patients, we found a septum bullosa in the site of contact. We defined septum bullosa as abnormal aeration of bony septum. It almost always involves perpendicular plate of ethmoidal bone. As the bone bifurcates, an air cell is formed inside the septal bone. The characteristics of pain in these cases were similar to patient with septal deviation. Anyway, this anatomical variation may be challenging during endoscopic surgery. In our experience, if there is convexity in both sides of a septum bullosa, it is better to dissect the mucoperichondrium in both sides and remove the whole septum bullosa along with its central chamber and mucus lining but if there is convexity only in one side, the lamella of the septum bullosa in the convex side can be removed and the mucus of the remaining lamella, will become in continuity with the mucus of the remaining septum.

In two cases, we found a medialized middle turbinate as the cause of the mucosal contact. In these cases, gentle manipulation of middle turbinate to make a space between middle turbinate and septum is necessary. Sometimes trimming of the most lateral part of middle turbinate tip should be performed to make a space for the newly repositioned middle turbinate.

Three patients had paradoxical curvature of middle turbinate. In a normal anatomy, the convexity of the middle turbinate is faced to the nasal septum while in a paradoxical curvature, this situation is contrary. All of these patients had also septal deviation and the involved turbinate was located next to the concaved side of the septum. Partial resection of middle turbinate was enough to make a distance between septum and the remaining of middle turbinate. We believe that complete resection of turbinate or any effort to completely correct the curvature in not necessary. A septoplasty is required in all cases.

Concha bullosa of middle turbinate (MTCB) was the most common abnormality in our patients and it also was the most known cause of RCPH. Pneumatization of the middle turbinate may originate from the agger nasi cells, frontal recess, sinus lateralis, posterior ethmoid cells, or directly from the middle meatus [1]. In our group, we found a great diversity in size and shape of MTCB. Most cases had MTCB in both sides. There was always a concomitant septal deviation. In most cases, the MTCB had one chamber but there were also cases with a two or three chambered concha bullosa. In order to remove the contact between septum and MTCB, we made a cut on the anterior face of the concha and resected the lateral lamella of concha bullosa and gently pushed the remaining of middle turbinate far from septum. In two cases with extremely large MTCB, we resected the medial lamella and medialized the remaining of middle turbinate to the extent that the new concha stayed in a reasonable distance from septum and the middle meatus was completely opened.

Bolger et al. [9] described different pneumatization types of middle turbinate and named them as lamellar type, bulbous type and extensive type of concha bullosa for pneumatization of vertical lamella, inferior segment and both the vertical lamella and inferior segment respectively. We considered pneumatization of vertical lamella as a separate category and named it lamella bullosa. We believe that both symptomatology and treatment of lamella bullosa is different from MTCB. Patients with lamella bullosa had a severe and durable pain in medial cantus and ipsilateral forehead while in MTCB pain was usually triggered by cold air or irritants and was not continuous in most of cases. In patients with a narrow lamella bullosa, a gentle lateralization of middle turbinate and medial lamella of lamella bullosa along with septoplasty will be sufficient. Resection of one of lamellas may weaken the insertion of middle turbinate to the extent that the turbinate becomes unstable and floppy so we recommend this operation only in carefully selected cases.

In 3 cases, superior turbinate was medialized to the extent that was stayed in a close contact with the upper parts of septum. To correct it, septoplasty with special care to the superior portions of the perpendicular plate of ethmoid bone and resection of superior turbinate were necessary.

Concha bullosa of superior turbinate (STCB) is difficult to find and is easily mistaken with a posterior ethmoidal cell. The best way to diagnose is looking for an elliptical air containing element in the region of superior turbinate that lays medial to the posterior ethmoidal cells. The medial lamella of CBST is attached to lateral limit of cribriform plate. Contact between upper septum and medial lamella of STCB is common. The symptoms of these patients were more severe than the ones with medialized superior turbinate. The patients with STCB feel pain and discomfort mainly in medial and lateral canthus and forehead. Surgical resection of medial lamella of CBST is theoretically sufficient for correction of mucosal contact but this procedure is technically difficult. Careful resection of whole superior turbinate is more practical and effective.

Bulla ethmoidalis is the largest cell in anterior ethmoidal region. When it is larger than usual, the medial wall of it may push the vertical lamella of middle turbinate medially and causes a contact with nasal septum. In rare cases, it may be large enough to contact with septum by itself. Large bulla ethmoidalis may also obstruct osteomeatal complex and predispose the patient to rhinosinusitis. To reverse this problem, an anterior ethmoidectomy and lateralization of middle turbinate is necessary. We found large bulla ethmoidalis as a cause for RCPH in 7 cases.

Contact between septum and ethmoid cells other that bulla ethmoidalis is also reported as a cause for RCPH [7]. We found two patients who had contact between medial wall of posterior ethmoidal cells and nasal septum. The posterior ethmoidal cell were hyperaerated and huge and this situation was bilateral in both cases. There was no concomitant septal deviation. A complete endoscopic ethmoidectomy with removal of medial wall of posterior ethmoidal cells was necessary to remove intranasal mucosal contacts in these patients.

We cannot opine about the exact mechanism for the differences in pain characteristics between various anatomical variations at this point. It seems that anatomical variations that make a larger contact area (e.g. lamella bullosa) and the ones with tighter contacts (e.g. septal spur) can lead to a more severe pain. Study of larger number of cases and a deeper look to the pathophysiology of this disease may be helpful to identify the exact mechanisms.

5. Conclusion

Multiple anatomical variations in nasal and paranasal sinuses may cause an intranasal mucosal contact and lead to rhinogenic contact point headache. Some of them, such as concha bullosa of middle turbinate are well recognized and common. We found 11 distinct anatomical conditions in patients with RCPH. All of them were surgically curable. The characteristics of pain may be different in each anatomical variation. Many clinicians are not familiar with the importance of some anatomical variations such as medialized turbinates, concha bullosa of superior turbinate and lamella bullosa. We showed that this variation deserve careful attention and must be addressed when a surgery for RCPH is planned.

Conflict of interest

None.

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