

CASE REPORT

A case of an uncommon anatomic variation of the middle turbinate associated with headache

Infrequente variazione anatomica del turbinato medio associata a cefalea

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SUMMARY

Headache due to the pressure of nasal mucosa in the absence of inflammation of the nose and paranasal sinuses is a clinical entity that has gained wide acceptance. Concha bullosa is the most commonly observed anatomical variation of the lateral nasal wall. The case is presented of a 31-year-old female with a history of intermittent frontal headache and bilateral nasal obstruction in whom we found the concha bullosa containing another, smaller concha bullosa inside. This is the first report of a case in which both outer and inner concha bullosa were septated (with two air cells inside). After resecting the lateral portion of outer concha bullosa and removing the inner concha bullosa, the patient reported no further headaches. The differential diagnosis of the variations of the middle turbinate and the relationships between the anatomic variations and pathophysiology of contact point headaches are discussed herewith.

KEY WORDS: Middle turbinate • Concha bullosa • Headache

RIASSUNTO

La cefalea correlata alla pressione della mucosa nasale in assenza di infiammazione del naso e dei seni paranasali è un'entità clinica ampiamente accettata. La conca bullosa è la variante anatomica della parete nasale laterale più frequentemente osservata. Viene qui riportato il caso di una donna di 31 anni affetta da cefalea frontale ricorrente e ostruzione nasale bilaterale in cui abbiamo riscontrato la presenza di una conca bullosa contenente una conca bullosa più piccola al suo interno. Questo è il primo caso riportato in cui le due conche bullose, interna ed esterna, erano separate, con due cavità aeree al loro interno. Dopo l'asportazione della parete laterale della conca bullosa esterna e la rimozione della conca bullosa interna, la paziente non ha più riferito cefalea. Vengono discusse la diagnosi differenziale delle varianti anatomiche del turbinato medio e le relazioni tra varianti anatomiche e punti di contatto scatenanti la cefalea.

PAROLE CHIAVE: Turbinato medio • Conca bullosa • Cefalea

Acta Otorhinolaryngol Ital 2009;29: EPUB 19 January

Introduction

Middle turbinate is an important landmark which forms the medial wall of the ethmoid sinus. It is associated with many functions of the nasal cavity, including humidification, lubrication of the upper airways, regulation of airflow and temperature, olfaction and filtration¹. Different types of middle turbinates, including pneumatized, paradoxically curved, bifurcate, trifurcate, secondary and accessory, have been described²⁻⁴, but pneumatized middle turbinate (concha bullosa) is the most common².

Chronic rhinosinusitis is well known for causing referred headache and facial pain over the area of the affected sinus and may be associated with relevant nasal and sinus symptoms, such as nasal obstruction, post-nasal discharge, purulent rhinorrhoea, and hyposmia⁵. However, many Authors have speculated that, even in the absence of inflammation of the paranasal sinuses, the referred head-

ache often results from pressure on the nasal mucosa due to anatomical variations⁶⁻¹⁰.

We present the first case of a female with a septated concha bullosa which contained a smaller septated concha bullosa within. This unusual anatomical abnormality was associated with headaches, probably caused by mucosal contact between the walls of the outer and inner concha bullosa.

Case report

A 31-year-old Caucasian female presented with a 5-year history of intermittent left-sided frontal and peri-orbital headaches, pain over the left cheek and bilateral severe nasal obstruction. She denied rhinorrhoea, epistaxis or post-nasal discharge, but noted that headaches lasted several hours and usually occurred followed by increasing left-sided nasal obstruction. About 15 years before our examination, she had been involved in a traffic accident with

severe injuries to the maxillofacial region, nasal pyramid and bony septum. Ten years after facial reconstruction and repositioning of the nose, she had the first episode of headache and facial pain. She was examined by a neurosurgeon, an ophthalmologist and an otorhinolaryngologist and was treated with non-steroidal anti-inflammatory drugs, topical corticosteroids and decongestants without success. The patient did not present any associated auras, photophobia, nausea, increased lacrimation or salivation. She was treated for migraine without aura by a neurologist, but no medications were effective.

At the time of presentation, anterior rhinoscopy demonstrated a septal deformation with right-sided spur in the region of the inferior turbinate. Nasal endoscopy showed healthy mucosa and hypertrophy of the left middle turbinate. Computed tomography (CT) scan of the paranasal sinuses was performed. The coronal CT plane demonstrated left-sided extensive middle turbinate pneumatization. The concha bullosa was septated. Axial CT plane showed septated concha bullosa with a bony formation inside the anterior air cell (Fig. 1). The next cross section showed the egg-shaped bony formation inside the concha bullosa cavity on the coronal plain (Fig. 2a). A pneumatized and septated bony formation inside the concha bullosa was found on the axial plane. It was found to be a septated concha bullosa within another, bigger septated concha bullosa. The anterior cell of the outer concha bullosa had a lateral ostium communicating with the middle meatus (Fig. 2b). Endoscopic examination showed no signs of sinus disease. By means of CT scan, we could find only one retention cyst inside the left maxillary sinus.

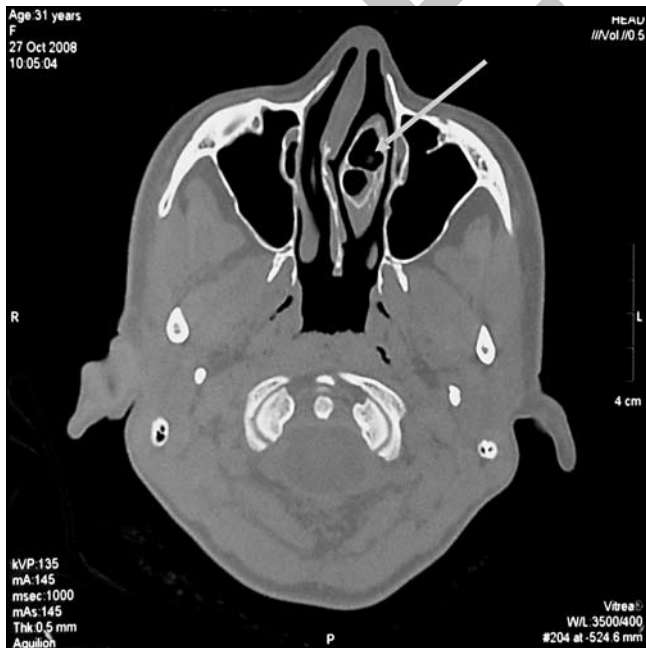


Fig. 1. Axial CT scan showing small bony formation inside anterior air cell of septated concha bullosa.

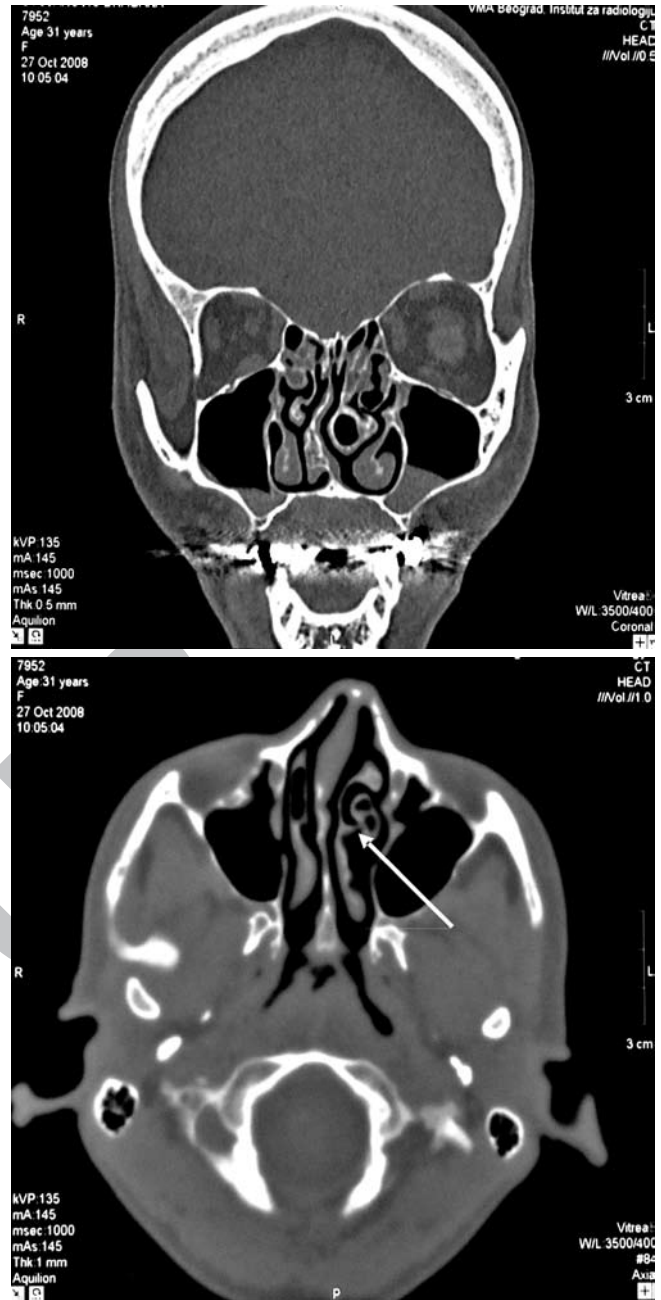


Fig. 2. a) Coronal CT scan demonstrating egg-shaped bony formation inside concha bullosa cavity. b) Axial CT scan showing septated inner concha bullosa in contact with outer concha bullosa wall (arrow).

The patient underwent transnasal endoscopic surgery under general anaesthesia. A vertical incision was made on the midline of the anterior wall of the outer concha bullosa and the lateral lamella was removed. The inner concha bullosa arising from the inside, as an egg-shaped mass with normal mucosa (Fig. 3), was also removed. The mucosa of the left-sided uncinate process and ethmoid bulla was also healthy. Finally, we performed septoplasty according to the Cottle procedure. Two days after the surgical treatment, the patient denied headaches and facial pain. She started to use saline nasal spray in each nostril for one month, and at 5 months' follow-up, reported no further headaches. Nasal

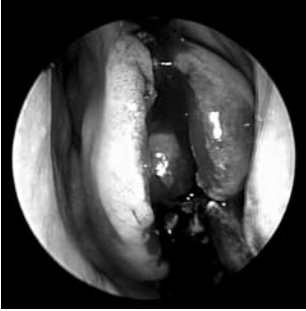


Fig. 3. Endoscopic view of inner concha bullosa after separation of medial and lateral portion of outer concha bullosa wall. Normal mucosa of inner concha bullosa can be seen.

breathing significantly improved after surgery. At the endoscopic nasal examination, 5 months after the surgical treatment, we found an epithelialized remnant of the outer concha bullosa, without middle meatal adhesions.

Discussion

Nasal turbinates are embryologically derived from a series of outgrowths

from the foetal lateral nasal wall. These outgrowths form a series of ridges referred to as “ethmoturbinals”². The middle turbinate develops from the third ridge and the superior turbinate develops from the fourth one². These variations in pneumatization of the “ethmoturbinals” may lead to anatomical variations within the bony structures of the ethmoidal complex². Partial or total pneumatization of the middle turbinate is called concha bullosa. This pneumatization may originate from the frontal recess, *sinus lateralis*, agger nasi cells, posterior ethmoid cells, or directly from the middle meatus². The degree of aeration may be variable. Bolger et al.¹¹ have found pneumatization of the vertical lamella of the middle turbinate (lamellar type), of the inferior segment (bulbous type), and of both the vertical lamella and inferior segment (extensive type). The exact reason for middle turbinate pneumatization is still unknown. Traumas, including intra-uterine foetal, peri-natal, and following delivery, developmental defects, growth anomalies of the maxilla and other facial structures, congenital deformities, shedding of incisors, breathing through the mouth are among the causes of nasal septal deviation¹². We assume that the trauma and developmental anomalies are also important factors in pneumatization of the middle turbinate. Microfractures in the bone of the middle concha could be sites for ingrowths of the nasal mucosa which is the precondition for spreading to the pneumatic space. In the case of our patient, some additional traumas, which were diagnosed during late puberty, could be the cause which contributed to the development of such an unusual form of pneumatization.

This is the second case to be reported of a patient with concha bullosa containing another concha bullosa inside. The first one was reported by Ozgursoy and Kucuk¹³. These Authors described a unique anatomical variant of the middle turbinate, a giant concha bullosa containing another large concha bullosa. However, our study is the first case to be presented in which both the outer and the inner concha bullosa were septated. Here, it could be due to two different sources of pneumatization in each of the middle turbinates, resulting in two air cells.

In the differential diagnosis, referring to our case, it should be noted that, in theory, there are pneumatized accessory and secondary middle turbinates. The accessory middle turbinate is defined as a medially inclined uncinat process which may give the impression that two middle turbinates are presented⁴. In our patient, however, we found a normal uncinat process in the left middle meatus. Secondary middle turbinate is a bony projection covered by soft tissue that originates from the lateral wall of the middle meatus^{4,14,15}. All cases reported in the literature were bilateral and none were pneumatized^{4,14,15}, therefore, we could not conclude whether inner concha bullosa was a pneumatized secondary middle turbinate.

The main symptom, in our patient, was intermittent frontal and periorbital headache. Wolff¹⁶ first described the concept of referred contact headaches occurring due to pressure points within the nose and suggested that headaches and facial pain could occur due to contact between the turbinate and other regions of the nasal cavity. Wolff's results showed that stimulation of various intra-nasal mucosal regions caused pain which was felt in the cutaneous distribution of the ophthalmic (V1) or maxillary (V2) division of the trigeminal nerve. Stammberger and Wolf⁵ found that mucosal contact could cause headache via substance P (SP), a neuropeptide released from the nasal mucosa. The receptors may be stimulated by chemical, caloric and also mechanical irritants, such as pressure. Stimulation of these polymodal receptors may cause SP release via both a central orthodromic impulse and a peripheral local, antidromic impulse. Locally, SP may cause vasodilatation, plasma extravasation, hypersecretion, and smooth muscle contraction. Release of SP in the central nervous system may cause referred pain⁵. Branches of the trigeminal nerve innervate the dura and intra-cranial blood vessels. When SP is released at peri-vascular sites, vasodilatation, plasma extravasation and peri-vascular inflammation can cause headache similar to migraine without aura⁸. Stammberger and Wolf measured the concentration of SP in human nasal mucosa using the radio-immunoassay technique and found that normal mucosa has higher concentrations of SP than chronic hyperplastic mucosa or polyp tissue⁵. In theory, contact between mucosal surfaces in the nose would elicit more pain than chronic infection or inflammation⁵. On the other hand, the nasal mucosa contains neural endopeptidase (NEP), an enzyme that degrades SP⁸. However, this degrading action of NEP may be disturbed by limited areas of mucosal contact where the epithelium is affected and the basement membrane (which contains a larger number of afferent nerves) is exposed⁸.

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 by a neurologist, ophthalmologist, internist and dentist to exclude other causes of headache, such as

neuralgia, temporal arteritis, vascular headaches or ischaemia^{6,9}. Evaluation for intra-nasal contact points should be included. The combination of CT scan with nasal endoscopy provides the maximum information⁶⁻¹⁰.

Wolff¹⁶ found that headaches may occur due to contact between the middle concha and septum or lateral nasal wall. Goldsmith et al.⁶ and Anselmo-Lima et al.⁷ called this phenomenon *Middle turbinate headache syndrome*. Behin et al.⁹ speculated that contact point headaches may be caused by the contact between the septum and the superior turbinate or medial wall of the ethmoid sinus and

not the middle turbinate. Huang et al.¹⁰ demonstrated that non-sinusitis related rhinogenic headache can be significantly reduced with surgical management following precise identification of the mucosal contact area. In our case, we found only one contact area, using CT scan and nasal endoscopy, between the inner concha bullosa wall and the bony septum of the outer concha bullosa. Only a few days after the endoscopic resection of the lateral portion of the outer concha bullosa and removal of the entire inner concha bullosa, headache significantly decreased and nasal breathing was significantly improved.

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Received: March 30, 2009 - Accepted: April 27, 2009