

# Periorbital Pain Induced by the Concha Bullosa Fungus Ball: A Case Report

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Concha bullosa is characterized by pneumatization of the middle turbinate and is one of the most common variations of the sinonasal anatomy. It is most often asymptomatic. A fungus ball in the concha bullosa is extremely rare, with only two reported cases in the English literature. This article describes a patient with such a fungus ball in the concha bullosa that caused right periorbital pain, specifically in the right medial canthal area, similar to mucosal contact point headache. *J Oral Facial Pain Headache* 2014;28:277-279. doi: 10.11607/ofph.1115

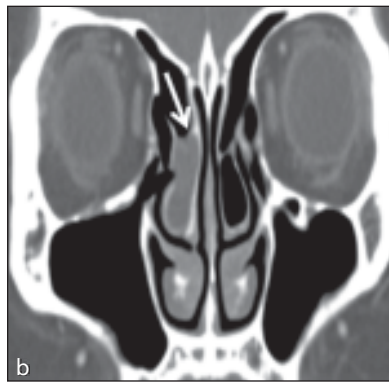
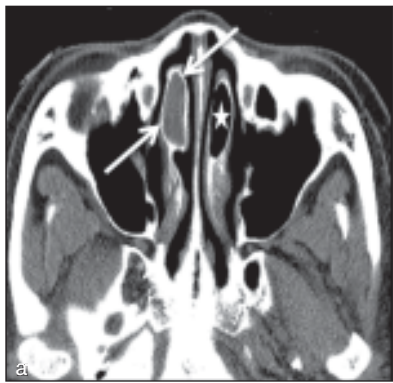
**Key words:** concha bullosa, fungus ball, periorbital pain, referred pain

Concha bullosa is characterized by pneumatization of the middle turbinate and is one of the most common variations of the sinonasal anatomy<sup>1</sup>; it can be unilateral or bilateral.<sup>2</sup> The incidence of concha bullosa ranges from 13% to 53%. This anatomic variation can be asymptomatic or symptomatic. For instance, a large concha bullosa may impair ventilation and drainage of the ostiomeatal complex and lead to sinus pathologies,<sup>2</sup> or cause a pyocele.<sup>3</sup> It can also produce headache or periorbital pain, also called middle turbinate headache syndrome, due to the middle turbinate compression against the septum or the lateral wall of the nose.<sup>4</sup> The pain is intermittent and localized to the periorbital and medial canthal or temporozygomatic region.<sup>5</sup> It is similar to mucosal contact point headache, which is a new entry in the International Classification of Headache Disorders (ICHD-2), for which evidence is limited.

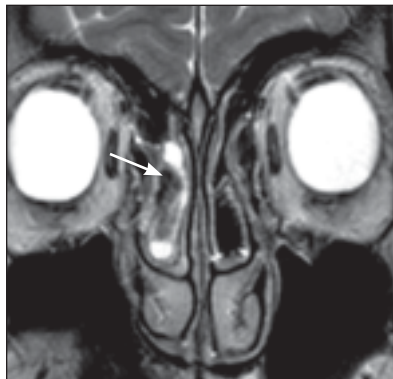
A fungus ball in a concha bullosa is extremely rare, with only two reported cases in the English literature.<sup>6,7</sup> In this article, an unusual case of a patient with the complaint of right periorbital pain, specifically in the right medial canthal area, caused by a fungus ball in the concha bullosa, is reported.

## Case Report

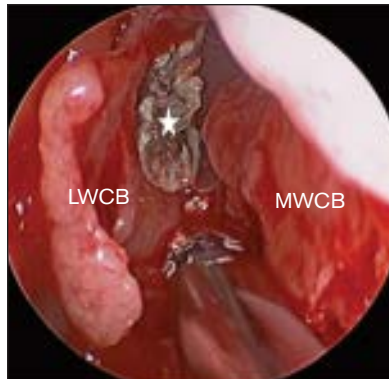
A 60-year-old woman presented with a 4-month history of continuous periorbital pain, specifically in the medial canthal area of the right eye. The character of the pain was atypical, dull, and pressure-like. There were no precipitating or aggravating factors. She had no history of ocular or nasal problems. There was no history of trauma or any significant past medical history. She first went to an ophthalmologist; funduscopy, analysis of eye movements, and neurologic examination revealed no abnormalities. Therefore, computed tomography (CT) of the orbit and a magnetic resonance imaging (MRI) of the brain were requested. The CT scans revealed a soft tissue shadow within the concha bullosa of the right middle turbinate (Fig 1). The MRI scans did not reveal intracerebral structural lesions but showed on the T1-weighted images an area of low signal intensity within the right concha bullosa that on the T-2 images appeared as a signal void (Fig 2). The differential diagnoses included polyp formation, submucosal cysts, cholesteatoma, ossifying fibromas, pyoceles, and tumors. Based on this finding, the patient was referred to the authors' department for further diagnosis and treatment.



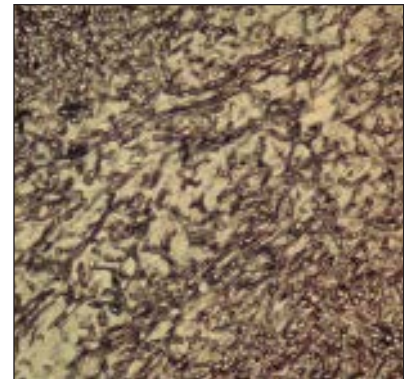
**Fig 1** CT scans—(a) axial view, (b) coronal view—reveal a soft tissue shadow within the concha bullosa of the right middle turbinate. The bony wall (a: arrows) of the concha bullosa of the right side is thicker compared to the contralateral side (a: star). The ostium (b: arrow) of the concha bullosa appears obstructed.



**Fig 2** T2-weighted MRI shows a signal void within the concha bullosa (arrow).



**Fig 3** Endoscopic view of the fungus ball mass (star). LWCB = lateral wall of the concha bullosa; MWCB = medial wall of the concha bullosa.



**Fig 4** Septated hyphae with 45-degree-angled branch are observed, consistent with aspergillosis (GMS stain × 400).

A sinonasal endoscopic examination did not reveal either a mucous/abnormal secretion in the nasal cavity or a deformity/displacement of anatomical structure in the nasal cavity. Under local anesthesia, a vertical midline incision was made in the anterior part of the middle turbinate, and the lateral portion of the concha bullosa was removed together with the edematous mucosa; this revealed a yellowish brown, cheese-like material that was suctioned (Fig 3). The histopathologic examination revealed the presence of aspergillus. This led to the diagnosis of a fungus ball or aspergilloma (Fig 4).

The postoperative course was unremarkable, and the patient was discharged 2 days later. She reported prompt pain relief and at a 12-month follow-up she was free of the pain with no evidence of recurrence.

## Discussion

Aspergillus is the most common reported cause of fungus ball; it predominantly involves the maxillary sinus without invading or penetrating the sinus mucosa, the associated blood vessels, and bone.<sup>8</sup> Patients suffering from fungus ball are usually immunocom-

petent. In the present case, a diagnosis of “concha bullosa fungus ball” (CBFB) was proposed, in accordance with the clinicopathologic criteria proposed by de Shazo,<sup>9</sup> because the aspergilloma was localized within the concha bullosa. To the best of the authors’ knowledge, this is the third case reported in the literature of CBFB presenting with periorbital pain.

The etiology of fungal infections in the paranasal region is unclear. One of the causes discussed in the literature is hypoventilation that could trap the fungal spores and provide anaerobic conditions for fungus ball development.<sup>10</sup> Concha bullosa is caused by anterior ethmoidal air cell migration into the middle turbinate and has a mucociliary transport system that connects the aerated cell to the frontal recess.<sup>3</sup> Therefore, the obstruction of the ostium and inflammatory changes within a concha bullosa might have led to mycotic infection of the patient. Indeed, the ostium of concha bullosa appeared obstructed in the CT images. However, Tsai et al<sup>11</sup> provided evidence that maxillary fungus ball is not associated with an obstruction of the ostiomeatal complex and that another as-yet-unexplained mechanism must be responsible. Recently, it has been hypothesized that mucosal immunity may play an important role in paranasal fungus ball development.<sup>12</sup>

The character of pain in the present patient was similar to mucosal contact point headache except for its duration. The pain was localized to the periorbital region, specifically the medial canthal area; however, the pain was continuous and not intermittent as it is in mucosal contact point headache, which is due to normal variations in mucosal edema in the nasal cycle and may be associated with a sensation of nasal congestion.<sup>4</sup>

The prompt disappearance of the periorbital pain after the CBFB removal is indirect proof that the pain was caused by the mycotic infection. However, the exact mechanism of the periorbital pain and the role of fungus ball in pain generation are still unknown. One possibility is referred pain. The anterior craniofacial structures are innervated by branches of the trigeminal nerve, and stimulation of various intranasal mucosal regions may cause pain that is felt in the cutaneous distribution of the ophthalmic (V1) or maxillary (V2) division.<sup>13</sup> In the present case, the stimulation of the anterior ethmoidal nerve that supplies the concha bullosa mucosa may have induced referred pain in the medial canthal area.

Another possibility is that the fungus ball may cause mucosal inflammation (that leads to headache or pain) by three possible mechanisms. First, the fungus ball may act as the site of bacterial infection, which causes acute bacterial mucosal inflammation. Second, obstruction of the natural ostium by the fungus ball itself may cause acute mucosal inflammation. Third, the fungus ball itself provides soluble antigens or irritants that may cause mucosal inflammation.<sup>14,15</sup>

Although the authors could not be sure that the concha bullosa fungus ball, because of its rarity, caused the periorbital pain, the concha bullosa fungus ball could be suspected on the basis of the CT and MRI findings. The characteristic findings of the fungus ball on CT are the heterogeneous opacification (microcalcification) and bony wall thickening, compared to the concha bullosa of the left side, and on MRI the hypointense signal on the T1-weighted image and the signal void on the T2-weighted image.<sup>16</sup> Surgery was both diagnostic and therapeutic in this case.

## Conclusions

A fungus ball in the concha bullosa may cause periorbital pain with characteristics similar to that of contact point headache.

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