

## Case Report

# Fungus ball in concha bullosa, an unusual site: a case report from hilly region

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### ABSTRACT

Fungus is a rare cause of rhinosinusitis in children. Its incidence has been reported as ranging between 13.5 and 28%. Fungus balls are extramucosal tangled masses of fungal hyphae. It commonly involves just one paranasal sinus. Most commonly maxillary sinus is involved and occasionally sphenoid sinus. Concha bullosa is one of the most common anatomical variations in the nasal cavity, with a frequency of reporting ranging from 14-53.6%. There are very few case reports in which Concha bullosa was affected with fungus ball. In this case report 13 years female child presented with complaints of swelling right cheek. The patient underwent endoscopic sinus surgery and there was involvement of concha bullosa and sphenoid sinus with fungus ball. Fungus was confirmed on microscopic examination and culture. Fungus ball is a rare presentation of fungal rhinosinusitis. It should be considered in enlarged middle turbinate with heterogenous opacity on CT. The diagnosis should be confirmed by microscopy or culture.

**Keywords:** Fungal ball, Concha bullosa, Rhinosinusitis, Endoscopic sinus surgery

### INTRODUCTION

Fungus is a rare cause of rhinosinusitis in children. Its incidence has been reported as ranging between 13.5 and 28%.<sup>1</sup> Depending on the presence or absence of tissue invasion, fungal diseases of paranasal sinuses are categorised into invasive and non-invasive. Fungus balls are extramucosal tangled masses of fungal hyphae.<sup>2</sup> It commonly involves just one paranasal sinus. Most commonly maxillary sinus is involved and occasionally sphenoid sinus. We are reporting a case of fungus ball in concha bullosa and sphenoid sinus, both of which are rare sites for its occurrence. Concha bullosa is the most common anatomical variation of nasal cavity. Its most common content is air. The presence of fungus ball in Concha bullosa is rare and only 9 cases have been published so far.

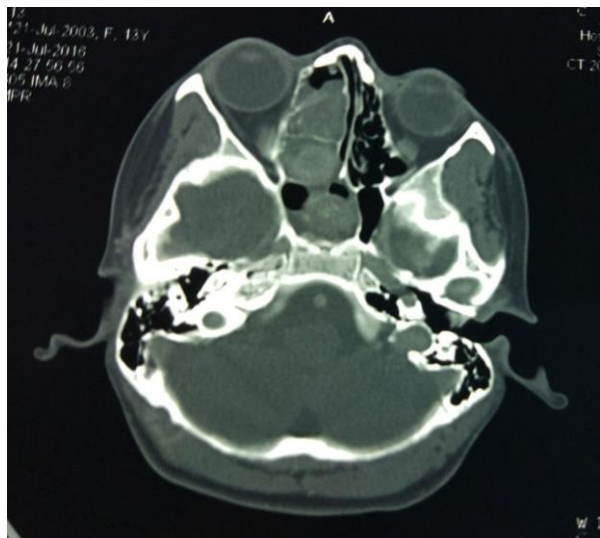
### CASE REPORT

A 13 year old female child presented to the Ear, Nose and Throat Outpatient department with complaints of swelling right cheek for the last 1 year. Swelling was insidious in onset and progressive in nature. It was not associated with any pain or headache or epistaxis.

She had undergone endoscopic sinus surgery for antrochoanal polyp one year back. Anterior rhinoscopy revealed an enlarged right middle turbinate with septal deviation towards left side and a greyish white polypoidal mass in right nasal cavity. There was discharge in the left nasal cavity. Clinical impression was of antrochoanal polyp.

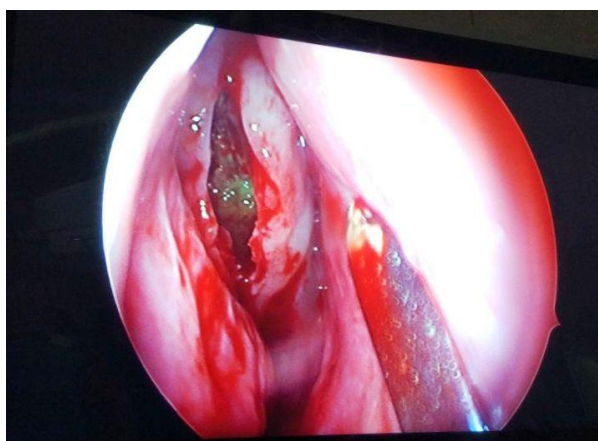
A paranasal computed tomography (CT) scan showed heterogenous soft tissue density with multiple focal areas

of hyperdensity causing complete opacification of right ethmoid and sphenoid sinus with extension to middle meatus and concha bullosa (Figure 1).



**Figure 1: CT scan showing complete opacification of right ethmoid and sphenoid sinus with extension to middle meatus and concha bullosa.**

During endoscopic sinus surgery right nasal cavity was visualised and concha bullosa was seen. On incising concha bullosa it was filled with yellowish brown coloured cheesy material (Figure 2). Ethmoidectomy and sphenoidotomy was done and it also showed the presence of yellowish brown cheesy material. Maxillary sinus was clear. The material removed was sent for microbiological examination and histopathological examination.

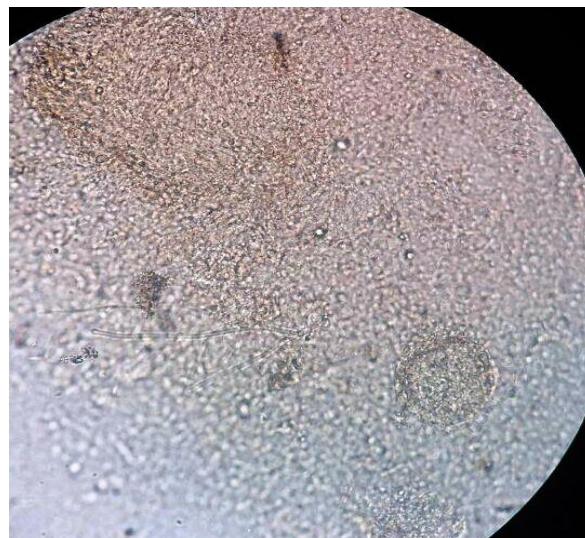


**Figure 2: Cheesy material seen during endoscopic sinus surgery in concha bullosa.**

In the microbiology laboratory, a 10% KOH mount was prepared from this cheesy material. It revealed the presence of hyaline fungal hyphae (Figure 3).

The tissue was minced and cultured on Sabouraud's dextrose agar media with and without cycloheximide at

25<sup>0</sup> C and 37<sup>0</sup> C. On this rapidly growing granular yellowish colonies appeared. On lactophenol cotton blue mount *Aspergillus flavus* was identified.



**Figure 3: KOH mount showing hyaline fungal hyphae.**

The patient was followed for 6 months during which she had no complaints and recurrence.

## DISCUSSION

Concha bullosa is one of the most common anatomical variations in the nasal cavity, with a frequency of reporting ranging from 14-53.6%.<sup>3</sup> It occurs when the pneumatization and enlargement of turbinates occur. It is most commonly seen in middle turbinate. It is pneumatized from frontal recess, agger nasi cells or middle meatus.<sup>4</sup>

It can be asymptomatic or present with headache, nasal obstruction or symptoms of sinusitis. The most common content of concha bullosa is air but rarely purulent material, bony septum, pyocele, ossifying fibroma, fungal mass and cholesteatoma have been found. In a retrospective study by Cucurova et al, of the 234 cases of concha bullosa operated only 31 (13.2%) had a pathologic entity. Fungus ball was present in one case (0.4%) only. So it is a rare entity and we have found only 9 case reports till now.<sup>5</sup>

Fungal rhinosinusitis is an uncommon condition. Its incidence has shown a marked increase in the recent years. This is due to improved diagnostic ability with wide use of CT in sinonasal pathologies and an increase in immunosuppressed patients.

Fungal rhinosinusitis occurs in two clinical forms as invasive and non-invasive. Non-invasive form is most prevalent. It includes allergic fungal sinusitis and fungal ball. Fungal balls are usually seen in immunocompetent persons. Common etiological agents of fungus ball includes: *Aspergillus fumigatus*, *Aspergillus flavus*,

*Pseudoallescheria boydii*, *Alternaria spp.* and other dematiaceous fungi.<sup>6</sup> Earlier fungus ball was known by the name of Aspergilloma, sinus aspergillois and mycetoma. It is usually seen in elderly females of 60- 70 years but there are case reports of pediatric age group involvement also. Agarwal has also reported two cases of paranasal sinus fungus ball in preadolescent age group.<sup>7,8</sup>

Our patient is also a female child of age 13 years. Fungus ball usually involves only one sinus. Most commonly it is maxillary sinus (94%), followed by sphenoid sinus (4-8%).<sup>8</sup> In another study by Alper et al from turkey fungus ball in maxillary sinus was seen in (77.7%) followed by (22.3%) in sphenoid sinus.<sup>9</sup> There are very few case reports in which concha bullosa was affected with fungus ball. In our patient there was involvement of concha bullosa and sphenoid sinus with fungus ball and both these are rare sites for this pathological entity.

CT is most reliable radiological investigation to identify a fungus ball.<sup>8</sup> Heterogenous opacification is the most common appearance associated with bony thickening or sclerosis of sinus wall involved. In present case heterogenous soft tissue density with multiple focal areas of hyperdensity were seen. The diagnosis of fungus ball is done on macroscopic and microscopic features. Macroscopically fungus balls are characterised by friable, cheese like material that can be green, yellow, brown or black and is easily peeled off the mucosa.

Microscopically there is an aggregate of tightly packed fungal hyphae. Positive fungal cultures are reported in 20-50% of patients. Though rare we could get the fungal growth in our patient.

In a case of fungus ball, surgery is always warranted. Nowadays endoscopic sinus surgery is done. This is generally curative and no local or systemic antifungal therapy is required.<sup>10</sup>

## CONCLUSION

Fungal ball is a rare presentation of fungal rhinosinusitis. Although it is very rare in concha bullosa and should be considered in enlarged middle turbinate with heterogenous opacity on CT. The diagnosis should be confirmed by microscopy or culture.

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