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Case Report

Fungal ball in concha bullosa as incidentaloma: A case report and the review of the literature

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ABSTRACT

Concha bullosa is an anatomic variant consisting in an enlargement and pneumatization of the middle nasal turbinate. A fungal ball (FB) localized in this structure is an extremely rare disease. This article describes the unusual case of a young patient with an asymptomatic fungal mass in the concha bullosa, incidentally discovered at computed tomography (CT) scan of the head, which was performed after trauma.

Introduction

The pneumatization of the middle turbinate, known as concha bullosa (CB), is a relatively common sinonasal anatomic variant, with an incidence ranging from 13% to 53% (Fig. 1) [1,2www].

CB usually becomes symptomatic when it is very large and causes compression of the nasal septum and obstructs drainage of the osteomeatal complex. Symptoms also arise from concurrent pathologies such as polyps, submucous cysts, ossifying fibromas, pyoceles or infections [3].

Fungal ball (FB) in CB is described as a non-invasive extramucosal overgrowth of hyphae. FBs are extremely rare in this anatomic site and generally characterized by nasal obstruction, fronto-orbital or retro-orbital headache, rhinorrhea, posterior discharge, and history of chronic sinusitis or recurrent rhinitis. FB can be unilateral or bilateral [1,4].

We describe the unusual case of a 12-year-old patient presenting FB in CB without reporting any symptoms,

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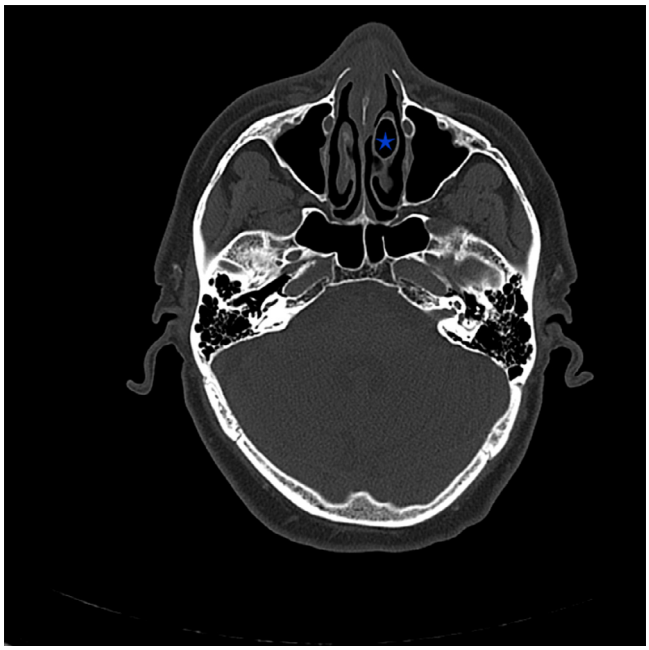

Fig. 1 – CT Axial plane, bony window. Left side middle turbinate pneumatization (star) in a patient without associated diseases.

incidentally discovered at computed tomography (CT) scan of the head performed after trauma.

As far as we know, this is the first asymptomatic pediatric case of fungus ball in concha bullosa, reported in the literature.

Case report

A 12-year-old girl was admitted to our hospital because of a mild head trauma after a car-vs-car collision. The patient complained of headache, mild dizziness, and loss of consciousness for a few seconds immediately following the trauma.

On physical examination, she was conscious (Glasgow Coma Scale:15) and she was breathing spontaneously, without visible lacerations over the scalp or skin bleeding.

No obvious changes of visual acuity and field were found. She had a history of thymoma, pollen allergy, and no history of previous trauma.

Routine laboratory tests were unremarkable.

A computed tomography (CT) scan of the head was performed to rule out acute subarachnoid or parenchymal hemorrhage and possible skull fractures. No skull fractures or intracranial bleedings were found.

Bilateral middle turbinate pneumatization was identified as an incidental finding. On the left side, the CB was enlarged, and filled with a high-density material containing coarse calcifications. Mucosal thickening was present.

Bony wall thickening with a mild rarefaction was noted. Opacification of some ethmoidal air cells was observed with no evidence of obstruction of the ipsilateral osteomeatal complex (*Fig. 2*).

The other paranasal sinuses were unremarkable.

The diagnosis of fungus ball involving concha bullosa was based on the imaging features described in literature [5–9] and the result of the rhino-endoscopic examination (*Fig. 3*) [5,6].

Since the case was discovered during the Covid-19 pandemic, the patient had no symptoms, and the surgery would have been demolitive, the multidisciplinary team decided for a clinical 6-month follow-up, unless symptoms would appear.

After 1 year the patient underwent a follow-up low-dose CT scan, showing no significant changes from previous images. The patient is still kept under observation through regular clinical follow-up.

Discussion

The origin of CB remains unknown. The hypotheses are different and some authors consider it to be a consequence of a developmental anomaly during intrauterine period [10,11].

Isolated fungus ball is a quite rare non-invasive fungal infection, mostly localized in the maxillary and sphenoidal sinuses. To the best of our knowledge, there are only 17 cases of FB in CB described in literature, including 2 cases reported by DuFour, but without any clinical or radiological informations. [1,3,6,8,10,12–21].

In almost all cases there is a clear female prevalence (86.6%) with a sex ratio of 13:2. The mean age of diagnosis is 36.4 years with extremes ranging from 10 to 88 years old.

The pathogenesis of the infection is still unclear. It may be caused by changes in ventilation due to the inflammatory obstruction of the ostium that connects the airy cell lumen of the CB to the frontal recess. This condition could trap fungal spores and provides optimal anaerobic environment for FB development. The most common infections are caused by Aspergillus sapp., especially A. fumigatus, and A. flavus [3,5,12,22].

Other known pathogens involved are Mucor spp., Candida, and Aureobasidium [8].

Currently recent studies have reported a relevant increase in the incidence of FB, probably due to the diagnostic technological improvement, such as CT imaging, as well as the progressive population aging [23,24].

Some predisposing conditions are known to encourage fungal infections such as diabetes, long-term antibiotic, chemotherapy, corticosteroid treatment, neoplastic conditions, and immunosuppressive diseases. However, mycotic infections of the paranasal cavity seem to be more common in otherwise healthy patients, without comorbidity or predisposing conditions [3,6,25].

All cases of FB in CB, described in literature, were symptomatic, complaining of nasal obstruction, headache, anosmia-hyposmia, retro-orbital pain, rhinorrhea, posterior discharge, chronic sinusitis or recurrent rhinitis [4,6].

The absence of symptoms has not yet been reported in any article, underling how rare our case is or probably how some cases go undetected because they remain clinically silent.

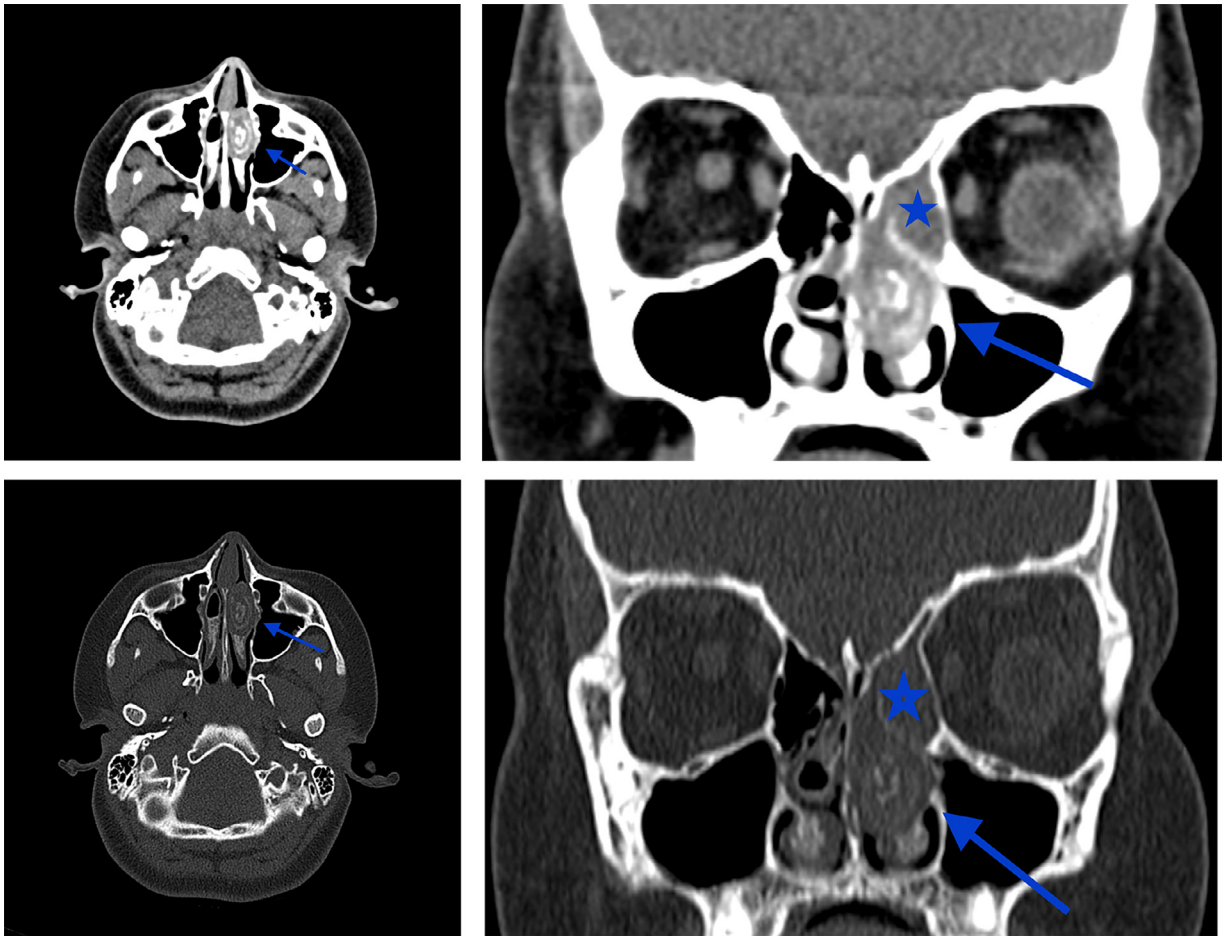

Fig. 2 – CT axial and coronal plane, soft tissue (A-B) and bony (C-D) windows. On the left side enlarged concha bullosa (arrow) containing hyperdense material and calcifications was found. Note the mucosal and bony wall thickening with rarefaction of the latter. Ethmoidal air cells opacification was associated (star).

Clinical examination itself is often inconclusive, with no specific findings such as middle turbinate hypertrophy, and septal deviation, probably due to the CB mass effect on the nasal septum [8].

Therefore, imaging tools are essential to reach a diagnosis.

CT scan is the best imaging modality for the radiological diagnosis of fungal infections of paranasal sinuses.

The main CT features described in literature are a complete or partial heterogeneous or homogenous opacification of the affected sinus, often with calcifications or microcalcifications, as a result of iron, calcium, and magnesium salts deposition detectable in its necrotic core. Areas of well-defined hyperdense foci are observed in 25%-50% of cases, strongly suggesting the diagnosis of a sinus fungus ball. [5,6,7,26]. In addition, other CT findings are cavity enlargement, mucosal thickening, bony sinus wall sclerosis, bony erosions, and irregular surface of the material, although mainly described in maxillary sinus fungus ball [1,3–7,10–15].

In particular both the erosion of the inner sinus wall and the irregular surface of the material seem to have high specificity and positive predictive value (PPV) for FB diagnosis, mainly in patients without intralesional hyperdensity [8,27]. In addition, irregular surface of the material can help to differentiate FB from chronic unilateral rhinosinusitis [27].

Recently advancement in imaging have led to an even better evaluation of the findings using more sophisticated tools such as CT volume rendering technique (VRT), which allows for a panoramic view of the involved region, providing further details, extremely useful for the pre-operative planning (*Fig. 4*) [28].

Magnetic Resonance Imaging (MRI) can also help to reach a diagnosis, but its long acquisition times, the high costs, as well as its lower availability limit its use in clinical practice. MRI is generally used as a second step, for assessment of complications such as bacterial superinfection, bacterial sinusitis, meningitis, and even brain empyema [8]. When MRI is performed, FB mass shows hyperintensity on T1-weighted images, and heterogeneous intensity on T2-weighted images, with central void signal. Hypertrophic mucosal walls in paranasal sinuses could be associated [8,29].

In all cases reported in the literature, CT scan was performed as first imaging technique [8] whereas only 3 patients underwent MRI, considered as a secondary imaging tool after CT scan.

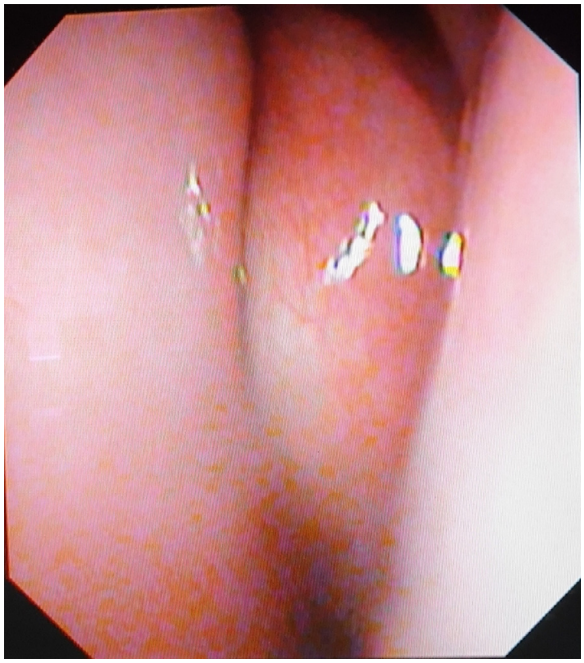

Fig. 3 – Rhinoscopic image showing concha bullosa.

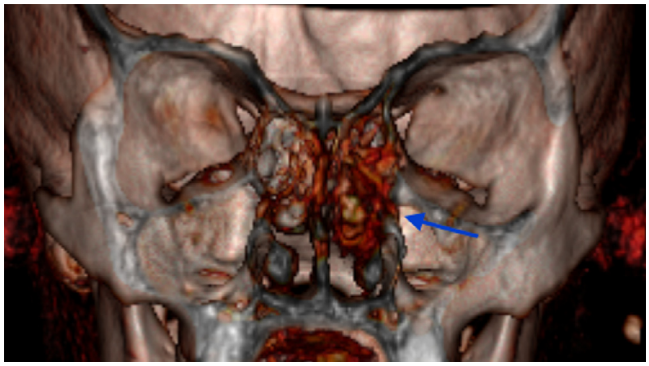

Fig. 4 – VRT reconstruction clearly shows mucosal and bony thickening with hyperdense material and calcifications inside the concha bullosa (arrow).

Differential diagnoses include bacterial sinusitis, mucocele, polyp formation, submucosal cysts, cholesteatoma, ossifying fibromas, pyoceles, inverted papilloma, squamous cell carcinoma or metastasis [1].

Definitive diagnosis is made mainly by the macroscopic appearance and histopathology, as cultures are negative in the 70% of cases, but imaging characteristics are typical [3,10,30], and CT findings could be used as diagnostic features in an appropriate clinical setting, avoiding invasive procedures.

Resolutive treatment of FB in CB is primarily based on endoscopic sinus surgery (ESS), consisting in the opening of CB, under local or general anesthesia [8].

Almost all patients described in literature underwent surgical treatment, except 1 patient who refused interventional management [8].

Treatment in asymptomatic patients is generally recommended as well; however, there is little evidence to support this approach [6].

The case we present here is the only known asymptomatic pediatric case, wherein the conservative approach has been proposed as the first line treatment.

In patients without symptoms and complications, especially if they are very young, we suggest clinical and imaging follow-up as first option - unless or until symptoms appear- because of the demolitive characteristic of surgical treatment.

Conclusion

The present case, to the best of our knowledge, is the first one that describes a patient with a fungus ball in concha bullosa discovered as incidentaloma. In an appropriate clinical setting CT findings could be used as diagnostic features, avoiding invasive procedures.

In young patients without symptoms or complications, conservative treatment, and imaging follow-up should be considered.

Patient consent

Written informed consent was obtained from patient's parents for the publication of this case report.

Conflict of Interest

All authors declare no conflict of interest and/or commercial involvement in the manuscript

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