

# Extraordinary pathologic entities within the concha bullosa

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## ABSTRACT

**الأهداف:** تحليل الأمراض الاستثنائية داخل الكونشا الفقاعية في دراسة استيعادية.

**الطريقة:** أجريت التداخلات الجراحية لعدد 136 مريض من (234 مريضاً مصاباً بالكونشا الفقاعية) بقسم الأنف والأذن والحنجرة وجراحة الرأس والعنق – مستشفى ازميز تيبك التعليمي ومركز الأبحاث – تركيا. في الفترة ما بين يناير 2002م حتى ديسمبر 2007م. تمت مراجعة النتائج الإشعاعية وأمراض الأنسجة لهذه الحالات في دراسة استيعادية.

**النتائج:** تمت فقط ملاحظة استرواح الأنسجة في التجويف لعدد 203 (86.8%) حالة من 234 حالة. تم ملء التجويف بالإفرازات القيحية قي 13 حالة (5.6%). كان الحاجز العظمي والقبيلة القيحية والسليلة المخاطية والورم الليفي العظمي والكرة الفطرية والورم الشحمي من الأمراض الاستثنائية الأخرى المصاحبة للكونشا الفقاعية.

**خاتمة:** على الرغم من أن حالات الكونشا الفقاعية لديها تجويف مستروح للأنسجة. إلا أنه ينبغي الأخذ بعين الاعتبار أن بعض الأمراض الاستثنائية يمكن ان تصاحب الكونشا الفقاعية.

**Objectives:** To retrospectively analyze the extraordinary pathologic entities inside the concha bullosa (CB).

**Methods:** Surgical interventions were performed on 136 patients (234 CB) at the Department of Ear, Nose, and Throat, and Head and Neck Surgery, Izmir Tepecik Training and Research Hospital, Izmir, Turkey between January 2002 and December 2007. Radiological and histopathological findings of these cases were reviewed retrospectively.

**Results:** In 203 (86.8%) of the 234 cases, only a pneumatized cavity was observed. The cavity was filled with purulent secretion in 13 (5.6%) cases. A bony septum, pyocele, polyp, ossifying fibroma, fungus ball, and cholesteatoma were the other extraordinary pathologies associated with CB.

**Conclusion:** Although most of the CB cases have pneumatized cavity, it should be kept in mind that some extraordinary pathologies can be associated with CB.

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Concha bullosa (CB) is the pneumatization of the concha, and is one of the most common variations of the sinonasal anatomy. A 14-53.6% frequency of CB was reported by various studies.<sup>1</sup> Pneumatization of the concha, regardless of the amount and the location, was defined as CB.<sup>2</sup> Bolger et al<sup>3</sup> classified pneumatization of the concha based on the location as lamellar CB, bulbous CB, and extensive CB.<sup>3</sup> The pneumatized cavity of the CB is lined by a respiratory epithelium like nasal cavity and paranasal sinuses. Like other aerated cells, the CB also possesses a mucociliary transport system.<sup>4</sup> Because of these features, the CB is susceptible to pathologic events like other aerated cells. However, pathologic entities arising from CB are uncommon, and there are only a small number of case reports in the literature on pathologic entities within CB. In this study, we reviewed 234 CB cases retrospectively, and reported the extraordinary pathologic entities arising from the CB.

**Methods.** This retrospective study was performed at Izmir Tepecik Training and Research Hospital,

Department of Ear, Nose, and Throat, Head, and Neck Surgery with ethical approval of the Hospital Research Ethics Committee. We searched our patient records for transnasal endoscopic middle turbinate surgery between January 2002 and December 2007. All of the operated CB cases were found and analyzed retrospectively, and all these cases were included in the study. Computed tomography (CT) scans, operation notes, and histopathological reports were reviewed. In some cases, the video-endoscopic operation records were also reviewed.

**Results.** We found 136 patients (234 CB) who were operated due to CB. Ninety-eight patients had bilateral CB. Ages of the patients were between 17 and 53 (mean 44) years. The surgeries of 16 patients were performed under general anesthesia, and the rest were performed under local anesthesia. When we reviewed the operation notes and CT scans for the 234 CB, we found that 203 (86.8%) of these were only pneumatized cavity. However, in 31 (13.2%) cases, there was a pathologic entity within the CB. The cavity was filled with purulent material in 13 (5.6%) of the cases, and bony septum (3%), pyocele (1.7%), polyp (1.7%), ossifying fibroma (0.4%), fungus ball (0.4%), and cholesteatoma (0.4%) were other pathologic entities seen within CB.

**Discussion.** Aeration and subsequent enlargement of the middle turbinate gives rise to the relatively common anatomic variant of CB. However, pathologic entities arising from the CB are uncommon. In 5.6% of our cases, purulent material was observed in the CB intraoperatively. Obstruction of the ostium is suspected to cause infection in the CB. It is believed that a CB usually drains directly into the ethmoid sinus.<sup>2</sup> Variations of this drainage pattern have been described.<sup>5</sup> When drainage and ventilation of the CB are disturbed, the CB might become infected. Lateral resection of the CB provided adequate drainage and ventilation in the cases of CB filled with pus.

Nasal polyps are the most common benign nasal mass and usually occur in the ethmoid sinus.<sup>2,6</sup> Reports of polyps arising in a CB have been infrequent in the literature.<sup>6</sup> Four (1.7%) of our cases have polyps in a CB. We do not know what caused polyp formation in our cases, but inflammation is suspected to promote such lesions. Resection of the polyp and the lateral portion of the CB was performed, and no recurrences were observed in these patients. In the literature, Yanagisawa et al<sup>7</sup> presented one septated CB case. In our study, we found 7 (3%) septated CB cases.

A mucocele is an epithelium-lined cavity containing mucus. Pyocele and empyema are synonyms describing an infected mucocele, with mucocele, and pyocele seen

mostly in the frontoethmoid complex of the paranasal sinuses.<sup>8</sup> A CB pyocele is rare and differs from an infected CB in its destructive potential and orbital invasion. In our retrospective analysis, we found 4 (1.7%) cases of CB pyocele. One of these is a large CB pyocele. Erosion of the lamina papyracea and destruction of adjacent structures is seen in **Figure 1**. During endoscopic biopsy, drainage of thick whitish pus was observed and diagnosis of pyocele was made. Aspiration of the pyocele combined with excision of the lateral part of the CB in this case and other CB pyocele cases. In all 4 cases, complete resolution was observed and no further treatment was needed.

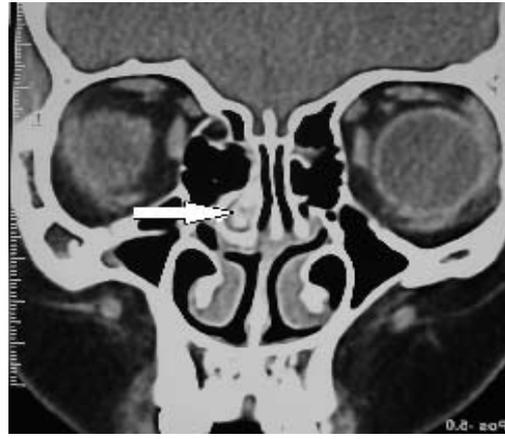
Ossifying fibroma, also a subgroup of the fibro-osseous lesions, is a benign osseous tumor with local aggressive behavior. The most common sites are the mandible and maxilla at craniofacial involvement. We found one case of ossifying fibroma in a CB, and there is no other case in the literature.<sup>9</sup> Radiographically, ossifying fibroma appears as an osseous lesion surrounded by unilocular, well-defined sclerotic border (**Figure 2**).<sup>10</sup> Ossifying fibroma can be seen as radiolucent, radiopaque, or mixed form radiographically depending on the amount of mineralized tissue that is present. Histopathologically, ossifying fibroma is composed of fibrous tissue that may vary in cellularity. The mineralized component may consist of plexiform bone, lamellar bone, and acellular mineralized material. In our case, the mineralized component is lamellar bone. Lamellar bone is surrounded by osteoblastic activity, and this feature is important in the differential diagnosis from fibrous dysplasia (**Figure 3**). Total excision is the treatment of this lesion.

In one of our cases, a fungus ball was detected inside the CB. In the paranasal sinuses, a fungus ball is usually found in just one sinus, most frequently the maxillary sinus, followed by the sphenoid sinus.<sup>11</sup> There is only one documented fungus ball of CB in the literature.<sup>12</sup> Our case is the second case in the literature. On CT scan of our case, hyperdense foci were observed inside the CB (**Figure 4**). Mucopurulent cheesy material was observed inside the CB intraoperatively, which almost totally filled the cavity. Histopathologically, non-specific chronic inflammation was observed in the mucosa. Mycelium with septate hyphae showing dichotomous branching was observed, and diagnosis of a fungus ball was confirmed (**Figure 5**).

Cholesteatoma is a relatively common disease within the middle ear cavity and temporal bone, whereas cholesteatoma of the paranasal sinuses is an exceptionally rare entity.<sup>13</sup> Our case is the first reported case of cholesteatoma inside the CB. Four theories on the pathogenesis of paranasal sinus cholesteatoma have been presented. The most accepted theory is



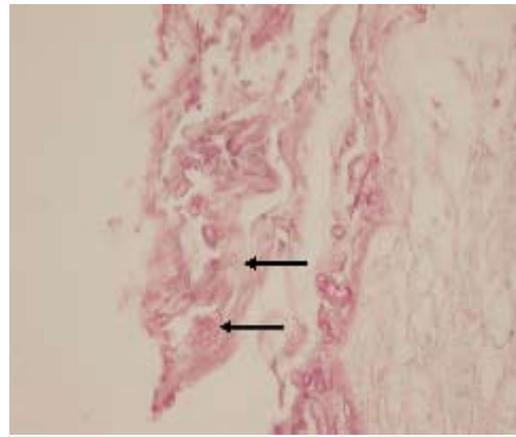
**Figure 1** - The coronal CT scan of concha bullosa pyocoele; homogenous fluid or soft tissue density lesion surrounded by a thin bony shell in the right nasal cavity (P), erosion of lamina papyracea (arrows).



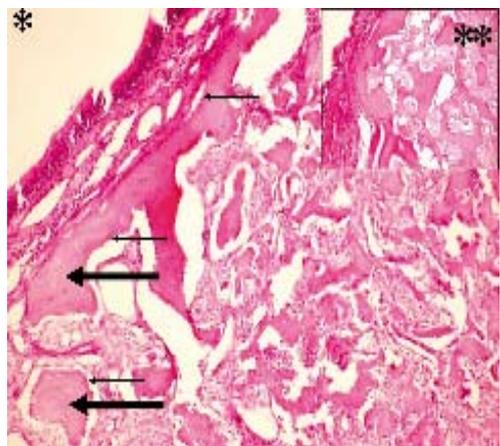
**Figure 4** - The coronal scan of fungus ball of the CB: hyperdense foci are observed in the concha bullosa (arrow).



**Figure 2** - The coronal CT scan of the ossifying fibroma inside the concha bullosa; intraosseous lesion surrounded by unilocal, well-defined sclerotic border (arrow).



**Figure 5** - The histopathology of the fungus ball of concha bullosa (Hematoxylin & eosin staining x220), non-specific chronic inflammation and mycelium with hyphae (arrows) are observed.



**Figure 3** - The histopathological section of the ossifying fibroma inside the concha bullosa (\*Hematoxylin and eosin staining x100, \*\*Hematoxylin and eosin staining x250), lamellar bone (thick arrows) is surrounded by osteoblastic activity (thin arrows).



**Figure 6** - Coronal CT scan of the patient with cholesteatoma inside the concha bullosa, erosion of lamina papyracea (small arrows) and ethmoid roof (big arrows) are observed.

congenital (primary) cholesteatoma theory. Congenital (primary) cholesteatoma is believed to be a result of misplaced ectodermal, epithelial cell remnants during the embryogenesis.<sup>14</sup> The development of acquired (secondary) cholesteatomas can be explained by 3 theories. One theory is the implantation theory that may occur after surgery or trauma. Immigration and metaplasia theories are the other theories. In our case, the congenital cholesteatoma theory is the highly accepted theory. The implantation theory cannot be accepted because our case has no history of previous trauma or surgery to the head and neck region. Immigration of epithelium from the nasal vestibule to the intranasal region has never been reported,<sup>13</sup> and we also could not show such an intranasal tract in our case. The metaplasia theory is rejected by many authors because squamous epithelium deriving from metaplasia due to chronic rhinosinusitis is of a non-keratinizing type.<sup>13</sup> In our case, CT scans showed a homogenous fluid or soft

tissue density lesion surrounded by a bony shell in the left nasal cavity. Erosion of the lamina papyracea and ethmoid roof was also determined on CT scans (Figure 6). Intraoperatively, when we removed the lateral part of the CB, the yellow-white colored mass was found at the posteroinferior part of the cavity. The mass was totally excised. Histopathological examination showed submucosal chronic inflammation, and squamous epithelium with keratinized debris and the diagnosis of cholesteatoma was made (Figures 7a & b). No recurrence of cholesteatoma was observed in the post-operative period.

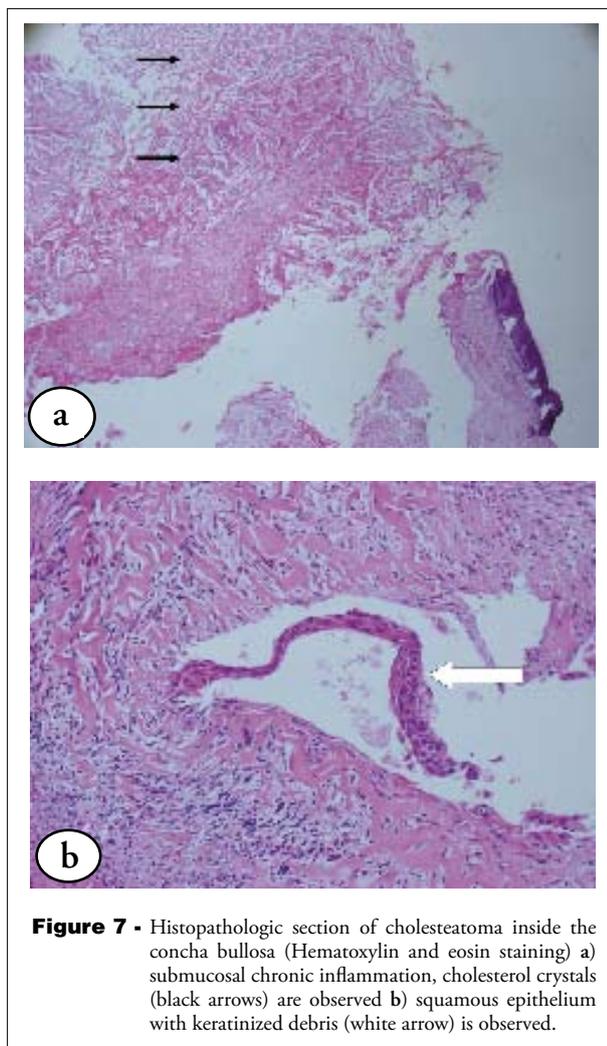
In this retrospective study, the analyzed CB cases were only operated CB cases. These cases do not reflect the whole population as there are also asymptomatic CB cases. Future studies must include the whole (operated and non-operated) CB cases for a more comprehensive analysis.

In conclusion, CB is the pneumatization of the concha, and is one of the most common variations of the sinonasal anatomy. Pathologic entities within the CB are uncommon, and there are only small numbers of case reports in the literature on pathologic entities within the CB. In our retrospective analysis of 234 CB, 86.8% cases had only pneumatized cavity, but 13.2% had extraordinary pathologic entities. Endoscopic endonasal surgery was performed on all the patients. Excision of the lateral part of the CB was performed in all cases. After total excision of the associated pathology, no recurrences were observed in the post-operative period. Although most of the CB cases have only a pneumatized cavity, it should be kept in mind that some extraordinary pathologic entities can be found within the CB.

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