

# Impact of Successful Choanal Atresia Repair on the Nasal Mucosa: A Preliminary Study

Ezzeddin Elsheikh<sup>1</sup> Mohammad Waheed El-Anwar<sup>1</sup> Hesham Radwan Abdel-aziz<sup>2</sup>

<sup>1</sup>Otorhinolaryngology-Head and Neck Surgery Department, Faculty of Medicine, Zagazig University, Egypt

<sup>2</sup>Pathology Department, Faculty of Medicine, Zagazig University, Egypt

Address for correspondence Mohammad Waheed El-Anwar, Otorhinolaryngology, Head and Neck Surgery Department, Faculty of Medicine, Zagazig University, Egypt (e-mail: mwenteg@yahoo.com).

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## Abstract

**Introduction** The main histological features of the nasal mucosa in choanal atresia are distorted cilia, marked increase of mucous submucosal glands associated with marked reduction of goblet cell density, and lymphocytic cellular infiltration.

**Objective** To study the nasal mucosal changes in cases of choanal atresia after successful repair compared with pre-repair mucosal histological features.

**Methods** Tissue samples were taken from the inferior turbinate of 3 patients (1 bilateral and 2 unilateral) who were successfully operated. Then, the biopsies were subjected to histopathological, histochemical and immunohistochemical studies. After that, the results were compared with pre-repair findings in the choanal atresia side and in the normal side.

**Results** Four biopsies (4 repaired choanal atresia sides) of the mucosa of the inferior turbinate revealed that 1 patient (who had a bilateral choanal atresia repaired), after achieving a patent choana for 8 months, had not completely recovered a normal nasal mucosa. The other 2 patients, after 18 and 23 months of achieving a patent choana, showed normal nasal cavities.

**Conclusion** The main histological features of the nasal mucosa in choanal atresia could be reversed by surgery, making the patients regain their choanal patency, with their mucosae changing back to normal gradually with time.

## Keywords

- ▶ choanal atresia
- ▶ nasal mucosa
- ▶ endoscopes

## Introduction

Congenital choanal atresia causes deprivation of nasal airflow and mucus transport,<sup>1,2</sup> and one of its features is accumulated tenacious mucous secretion in the nasal cavity.<sup>3</sup>

The main found histological features of the nasal mucosa in choanal atresia are distorted cilia,<sup>3,4</sup> marked increase of mucous submucosal glands associated with marked reduction of goblet cell density, and lymphocytic cellular infiltration.<sup>3</sup> This raised a question: are these found features permanent, and will these patients complain of thick mucus secretion for life? Or will these features reverse after successful surgery?

Therefore, the aim of this work was to investigate the impact of the successful permanent repair of the choanal atresia on these histological features.

## Patients and Methods

This study was conducted at the Otorhinolaryngology–Head and Neck Surgery and Pathology departments in the period between March 2011 and February 2015 on patients diagnosed with choanal atresia. Patients with systemic mucociliary diseases were excluded from the study.

The patients who needed general anesthesia for another surgical reason 6 months or more after the successful repair

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of the choanal atresia were the candidates in this study who allowed for the performance of a biopsy of their inferior turbinate mucosae. The repair was considered successful if the patient showed easy nasal breathing, non-interrupted oral feeding, a patent nasal airway, and an open new choana. Patency was defined as less than 50% restenosis.<sup>5-7</sup>

The patients were subjected to full history taking, general and local examinations, and routine preoperative laboratory tests. A written informed consent to participate in the study was signed by the relatives of the patients, and the university's institutional review board approved the study.

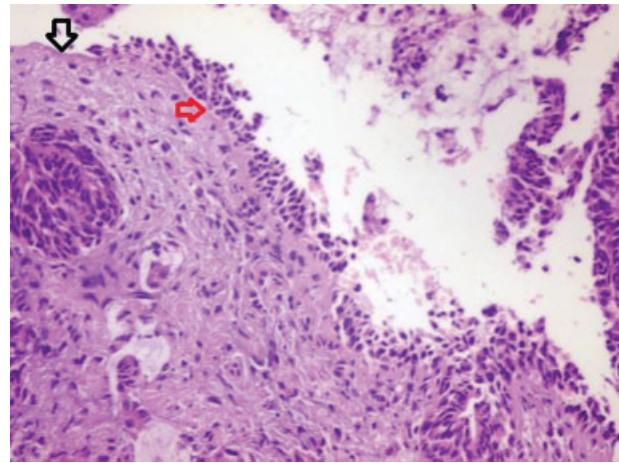
With the patients under general anesthesia, a biopsy of the inferior turbinate mucosa (0.5 cm behind its anterior end) was obtained. Then, the biopsies were subjected to histopathological, histochemical and immunohistochemical studies, as described by Elsheikh et al.<sup>4</sup>

**Results**

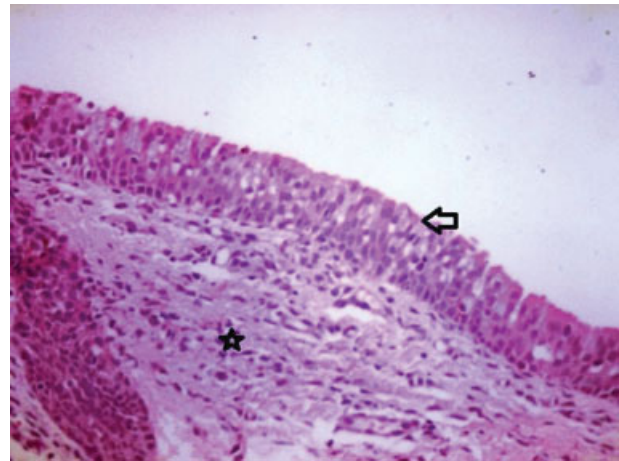
A total of three patients who were successfully operated for choanal atresia were included in this study (one was repaired for bilateral choanal atresia, and two for unilateral choanal atresia); their demographic data are summarized in **Table 1**. The repair was performed by stentless transnasal endoscopic approach, with resection of the posterior part of the vomer in all cases.

The histopathological study of the 4 biopsies of the mucosa of the inferior turbinate (4 repaired choanal atresia sides) revealed that in 1 patient (who had bilateral choanal atresia) after 8 months of repair, the findings were similar to the ones prior to the repair in the form of pseudostratified columnar ciliated covering epithelium with focal mucosal atrophy and ulceration, and with intact basement membrane (**Fig. 1**). But the mucosa had features unlike the ones found prior to the repair, because the basement membrane was of normal thickness. Moreover, the overall number of goblet cells increased significantly after the repair, to 17–21 cells per high power field (HPF). The submucosa also showed less lymphocytic infiltrate than before the repair (**Figs. 1 and 2**).

Compared with the findings prior to surgery, in the other 2 patients (2 unilateral choanal atresias), after 18 months and 23 months of the repair with patent choana, the covering epithelium was pseudostratified columnar ciliated epithelium with no mucosal atrophy or ulceration, and with an



**Fig. 1** Photomicrograph shows the pseudostratified columnar ciliated epithelium with focal mucosal atrophy and ulceration (black arrow) with an intact basement membrane (red arrow), H&E x 400.

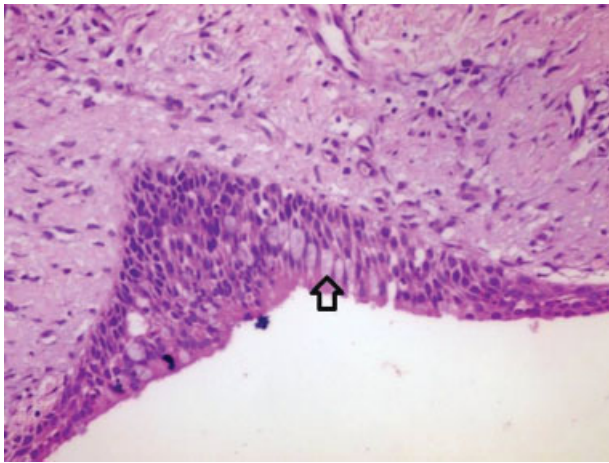


**Fig. 2** Photomicrograph showing an increased number of goblet cells (bold arrow) mild lymphocytic infiltrate in the submucosa (star), H&E x 400.

intact basement membrane of normal thickness. The number of goblet cells also increased, to 20–25 cells per HPF (**Fig. 3**). The submucosal glands were found in lower numbers and less active than before the repair, mainly of the mucous type

**Table 1** Patient data

Patients	Patient 1		Patient 2	Patient 3
	Atretic side 8 months after repair (patent)	Atretic side 8 months after repair (patent)	Atretic side 1.5 year after repair (patent)	Atretic side 23 months after repair (patent)
Gender	Male	Male	Male	Male
Age at the time of the biopsy	8 months	8 months	6.5 years	9 years
Side	Bilateral	Bilateral	Unilateral	Unilateral
Nature of the atresia	Pure bony	Pure bony	Mixed	Pure bony



**Fig. 3** In the second case (18 months after the repair), photomicrograph showing the pseudostratified columnar ciliated epithelium with no mucosal atrophy or ulceration, and with an intact basement membrane of normal thickness and more goblet cells (arrow).

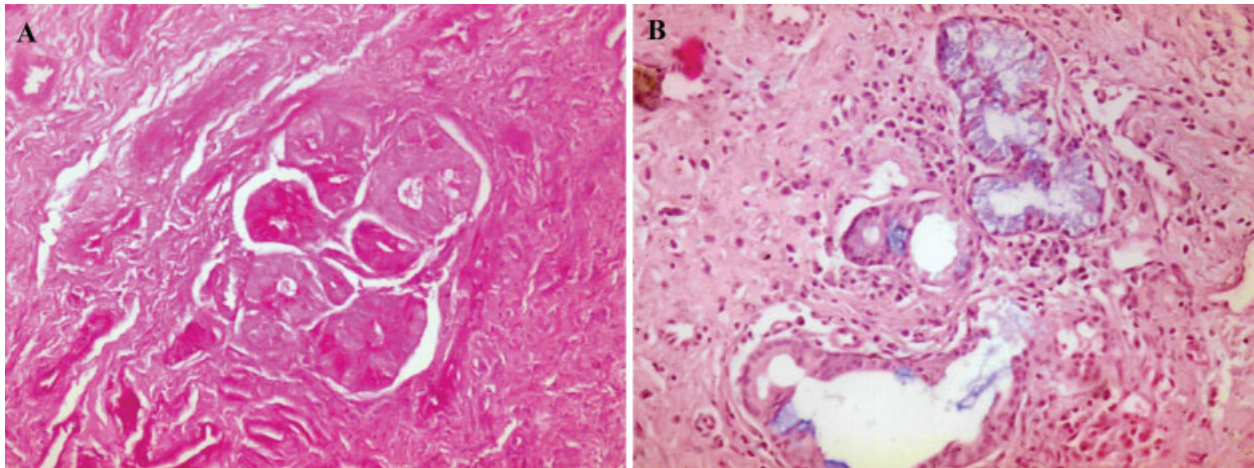
and showing faint periodic acid-Schiff (PAS) - positive, faint Alcian blue - periodic acid shift (PAS) positive material (**►Fig. 4**). The submucosa showed less lymphocytic infiltrate than before the repair (**►Figs. 3, 5**) (**►Table 2**).

Collectively in all biopsies, the mean of goblet cells was  $21.5 \pm 1.9$ , which was nearly similar to the levels of a normal mucosa.<sup>3</sup> On the immunohistochemical studies, the nerve terminals (using neuron specific enolase, NSE, immunostaining) were found to be intact. In addition, lymphocytic infiltrate proved by CD45 immunostaining (**►Fig. 5**).

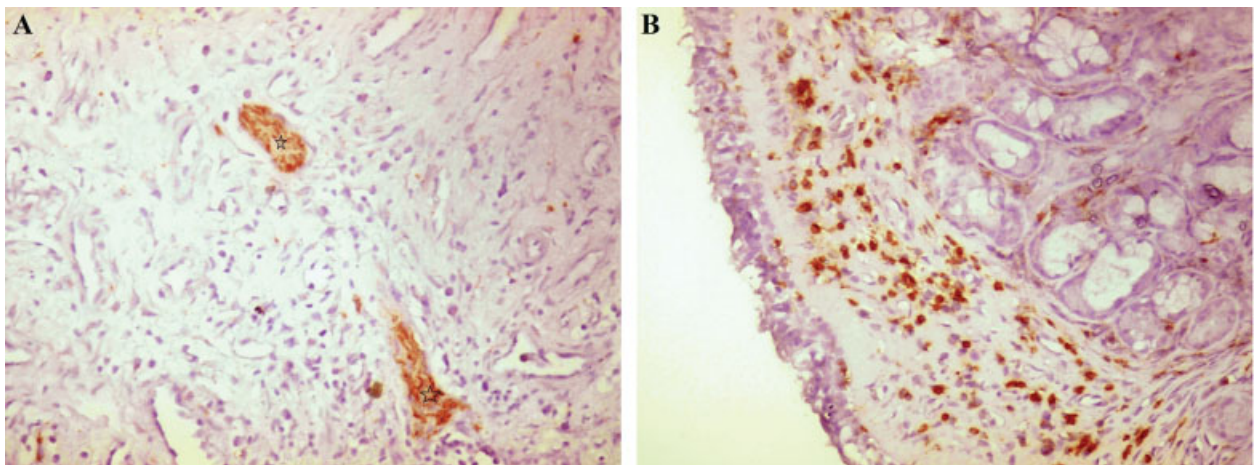
After 18 months of achieving a patent choana, the mucosa had the same features as it does in healthy subjects.

### Discussion

Basal, goblet and columnar cells are usually identified in the respiratory airway epithelium. Goblet cells are mucus producing cells with an important role in mucociliary



**Fig. 4** (A) Photomicrograph showing faint staining for neutral mucin, periodic acid-Schiff (PAS) X 400; (B) faint staining for acidic mucin, Alcian-PAS X 400.



**Fig. 5** (A) Immunostained for neuron specific enolase (NSE) showing intact wavy nerve bundles (star), (B) mild lymphocytic infiltrate, Hematoxylin counterstain X 400.

**Table 2** Changes in the histopathological results after the repair of the choanal atresia

Histopathological results	AS before surgery	AS after 8 months (patent)	AS after 8 months (patent but narrower)	AS after 1.5 year (patent) (unilateral, 6.5 years)	AS after 23 months (patent) (unilateral, 9 years)	Patent side (no atresia)
Epithelium	Focal mucosal atrophy and ulceration	Focal mucosal atrophy and ulceration	Focal mucosal atrophy and ulceration	No atrophy or ulceration	No atrophy or ulceration	No atrophy or ulceration
Basement membrane	Thickened	Normal thickness	Normal thickness	Normal thickness	Normal thickness	Normal thickness
Submucosal glands	Diffuse and marked increase in number	Moderate	Moderate	As normal	As normal	Less than the atretic side
Lymphocytic infiltrate	Moderate increase in lymphocytic infiltrate in the submucosa	Minimal	Minimal	Minimal	Minimal	Minimal

Abbreviation: AS, atretic side.

clearance and in the local mucosal immune system. Goblet cell density differs in various sites of the respiratory tract, and is affected by different factors, such as genetic factors and the impact of air currents, as well as infection or inflammation.<sup>8</sup>

The effect of airflow deprivation on the nasal mucous membrane is an issue of wide disagreement in the literature. In experimental animals, studies showed an increase in goblet cells in the closed side, and loss of cilia with squamous metaplasia.<sup>9,10</sup> Mogenson and Tos<sup>10</sup> detected an inverse relationship between the number of goblet cells and the air current, and found abundant goblet cells in regions with scanty airflow and vice versa. Contrary to that, Berger et al<sup>11</sup> detected more goblet cells in the lateral wall of the inferior turbinate compared with the medial wall, and She et al<sup>8</sup> reported more goblet cells in the lateral wall of the uncinate process compared with the medial wall, and this reduced number was attributed to epithelial changes caused by air currents.

A former study found that the nasal mucosa in the atresia side in both unilateral and bilateral choanal atresias is not histologically different, and is characterized by histological features such as thickened basement membrane, marked increase of mucous submucosal glands associated with marked reduction of goblet cell density, and lymphocytic cellular infiltration.<sup>3</sup> The choanal atresia side is also characterized by accumulated tenacious mucous secretion in the nasal cavity.<sup>4,12</sup>

Compared with serous glands, abundant submucosal active mucous glands increase mucous viscosity<sup>13</sup>, leading to stasis of the thick mucus as result of the low viscosity of the periciliary sol, which allows the cilia to beat and propel the mucous blanket to the mouth. The lack of mucous drainage could also be increased by the constricted glandular excretory ducts of the thick basal membrane that was detected in the nasal mucosa of the choanal atresia side.<sup>4</sup>

The current study aimed to evaluate the impact of the permanent repair of the choanal atresia on these histological features. We found that the mucosal features of choanal atresia reversed after achieving a permanent patent airway, and the mucosa became close to normal with more patency time. Thus, the histological features of the nasal mucosa associated with the choanal atresia are mostly secondary to complete nasal obstruction. Therefore, the successful repair of the choanal atresia not only recreates nasal airflow and breathing, but also restores the features of the normal nasal mucous membrane and the mucus gradually with time after achieving a persistent nasal patency.

The limitation of this preliminary work is the small number of biopsies, as it was difficult to find choanal atresia patients successfully operated who underwent surgery under general anesthesia for another reason and accepted to have a biopsy performed on them. However, this study opens the door for further studies on large series of patients, and for the comparison of the results in bilateral and unilateral choanal atresias.

## Conclusion

The main histological features of the nasal mucosa in choanal atresia could be reversed by surgery, with the patients regaining choanal patency, and the mucosa going back to normal gradually with time.

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The authors declare no financial support for this study.

### Conflicts of Interest

The authors have no conflicts of interest to declare.

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