

# Nasal Myoepithelioma Removed through Endonasal Endoscopic Surgery: A Case Report

Terumichi Fujikura<sup>1,2</sup> and Kimihiro Okubo<sup>1</sup>

<sup>1</sup>Department of Head & Neck and Sensory Organ Science, Graduate School of Medicine, Nippon Medical School

<sup>2</sup>Department of Otolaryngology, Tokyo Woman's Medical University Medical Center East

## Abstract

Myoepithelioma is a rare neoplasm that can occur in either the major or minor salivary gland and accounts for less than 1% of salivary gland neoplasms. We report a rare case of a nasal myoepithelioma that originated from the nasal inferior turbinate. The tumor, measuring 50 × 30 × 20 mm, was in the right nasal cavity and had a necrotic surface. We removed the tumor through endonasal endoscopic surgery. The tumor had spindle-shaped cells and was positive for cytokeratin, (AE1/AE3), vimentin, S-100β, and MIB-1 but was negative for CD34, desmin, neuron-specific enolase, and synaptophysin. Slight immunoreactivity for smooth muscle actin was noted in some tumor cells. There has been no evidence of tumor recurrence in the 18 months following surgery.

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**Key words:** myoepithelioma, nasal cavity, endoscopic surgery, cytokeratin

## Introduction

Myoepithelioma is a rare neoplasm that can occur in either the major or the minor salivary gland; however, it accounts for less than 1% of all salivary gland neoplasms<sup>1</sup>. To the best of our knowledge, there have been only 3 reports of nasal myoepithelioma in the English-language literature<sup>2–4</sup>. We report a case of nasal myoepithelioma and discuss its diagnosis, histological characteristics, and treatment.

## Case Report

A 72-year-old man presented with a 3-month

history of epistaxis and nasal obstruction. Physical examination revealed a large tumor with a necrotic surface which had originated from the nasal inferior turbinate in the right nasal cavity (**Fig. 1**). The lesion was firm and did not move easily with palpation. Contrast-enhanced coronal computed tomography (CT) scans were obtained. The CT scan showed a heterogeneous mass occupying the entire nasal chamber. Mild deviation of the nasal septum and widening of the right nasal cavity were seen. Osteolytic substances were not found in the nasal cavity or sinuses (**Fig. 2**). T2-weighted magnetic resonance imaging (MRI) showed high signal intensity in the tumor (**Fig. 3**). A careful endoscopic examination showed little space between the tumor and the nasal septum or the nasal turbinate.

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Correspondence to Terumichi Fujikura, Department of Otorhinolaryngology, Nippon Medical School, 1–1–5 Sendagi, Bunkyo-ku, Tokyo 113–8603, Japan

E-mail: teru-fujik@nms.ac.jp

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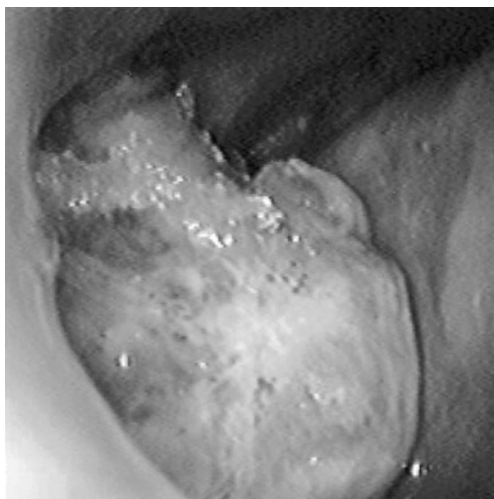


Fig. 1 The findings of endoscopic examination. We found a large tumor with a necrotic surface which had originated from the nasal inferior turbinate of the right nasal cavity.



Fig. 3 T2-weighted magnetic resonance imaging shows high signal intensity in the tumor.



Fig. 2 Coronal computed tomogram shows a heterogeneous mass occupying the entire chamber of the right nasal cavity.

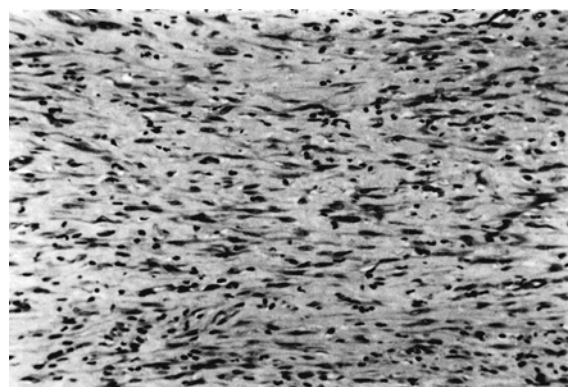


Fig. 4 Tumor section shows a myxoid pattern of growth with spindle-shaped cells (hematoxylin and eosin, original ×400).

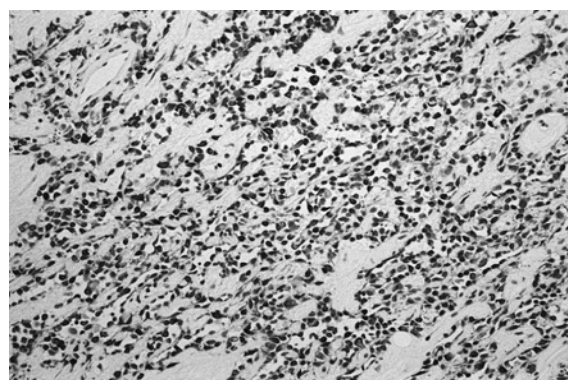


Fig. 5 Tumor section shows positive staining for cytokeratin (AE1/AE3, original ×200).

An excisional biopsy was performed from the nonnecrotic region of the tumor. The reactivity for MIB-1 was low grade. The tumor nuclei showed no atypical mitoses. Histological examination showed that the growth pattern of this tumor was myxoid with collagen fiber proliferation. Myxoid zones were composed of spindle-shaped cells (**Fig. 4**). Mitotic activity was absent. The biopsy specimen was positive for cytokeratin (AE1/AE3) (**Fig. 5**), vimentin, S-100 $\beta$ , and MIB-1 but was negative for CD 34, desmin, neuron-specific enolase, and synaptophysin. Slight immunoreactivity for smooth

muscle actin was noted in some tumor cells. A small number of duct-like structures were occasionally observed. The appearance of the mass on light microscopy and its pathologic, histological, and

immunohistochemical characteristics were consistent with myoepithelioma.

On the basis of these results, we performed endonasal endoscopic surgery with the patient under general anesthesia and excised the tumor totally. Through the enlarged endoscopic view, we could clearly identify the portions of the nasal cavity the tumor had invaded. Further endoscopic examination suggested that the tumor had originated from the inferior turbinate near the fontanelle but had not invaded the maxillary sinus. We also resected the inferior turbinate because it showed severe atrophic change due to tumor invasion. The tumor mass was partially adhesive to the nasal septum and the lower portion of the middle turbinate mucosa. Therefore, the mucosa of these regions was also resected. The tumor mass could be removed with minimal bleeding.

The tumor mass measured  $50 \times 30 \times 20$  mm and was not encapsulated. The results of immunohistochemical staining of the tumor were the same as those of the biopsy specimen.

### Discussion

The main symptoms of nasal myoepithelioma are rapid enlargement of the tumor mass and epistaxis<sup>2,4</sup>. In our case, epistaxis and nasal obstruction had been noted by the patient for 3 months, but there was no pain or other specific symptoms. The imaging appearance of a myoepithelioma is generally nonspecific;<sup>4</sup> therefore, preoperative pathologic and endoscopic examinations are decisive for therapeutic planning.

Primary pleomorphic adenomas of the nasal cavity account for approximately 18% of sinonasal nonepithelial neoplasms<sup>5,6</sup>. However, nasal myoepithelioma is a much rarer low-grade neoplasm. Nasal myoepitheliomas are composed of myoepithelial cells with solid, myxoid, or reticular patterns of growth. The cells themselves may be spindle-shaped, plasmacytoid, epithelioid, or of the clear-cell type<sup>7</sup>. Myoepithelioma has traditionally been defined as a tumor without ductal differentiation. However, some authors have adopted a less rigid definition to include the presence of a

small number of ducts<sup>2</sup>. Including the present case, 3 of the 4 reported nasal myoepitheliomas had a small number of ductal components<sup>2,4</sup>.

In the sinonasal setting, this type of rare neoplasm is difficult to distinguish from other soft-tissue tumors that show spindle cells or myxoid features. Spindle-cell myoepitheliomas must be distinguished from leiomyomas, leiomyosarcomas, nerve sheath tumors, and synovial sarcomas. Staining for cytokeratin is reported to be helpful for ruling out other mesenchymal neoplasms<sup>2,5</sup>. Staining for S-100 protein is also useful in some cases. In our patient, staining for cytokeratin and S-100 $\beta$  was positive. Strong, diffuse staining for cytokeratin was observed. On the other hand, the presence of chondroid tissue and ductal differentiation would support the diagnosis of pleomorphic adenoma. An infiltrative growth pattern, in contrast to well-circumscribed borders of benign neoplasms, is a characteristic of a myoepithelial carcinoma<sup>7</sup>.

In all reported cases of myoepithelioma, surgical treatment was performed. Partial maxillectomy via a lateral rhinotomy approach or a Caldwell-Luc procedure would be performed for patients who were suspected to have a low-grade sarcomatous neoplasm or for those who showing destruction of the posterior wall of maxillary sinus<sup>2,3</sup>. Another patient who underwent endonasal surgery had a tumor nearly 10 mm in diameter<sup>4</sup>. In our patient, the tumor mass occupied the entire right nasal cavity. However, on preoperative endoscopic examination little space was observed between the tumor and the nasal septum or the nasal turbinate. These findings suggested that the tumor could be removed with endoscopic surgery. On the basis of the pathological examination of the biopsy specimen we suspected the tumor was a benign myoepithelial neoplasm. The patient, who was 72 years old, preferred a minimally invasive operation. Therefore, we decided to perform endonasal endoscopic surgery. We were also prepared to change during the operation to a lateral rhinotomy approach. We could remove a relatively large tumor mass by means of endonasal endoscopic surgery, which caused minimal bleeding or scarring. The patient has been followed up for 18 months postoperatively

without macroscopic evidence of tumor recurrence. A careful endoscopic endonasal technique allows minimally invasive surgery for the treatment of massive benign tumors of the nasal cavity.

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