

Recurrent Myoepithelioma Treated by Palatal and Piriform Apertural Approaches

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Summary: Myoepithelioma is a rare, benign salivary neoplasm, most frequently located in the salivary gland; extrasalivary cases most commonly occur in the palate. This tumor is prone to recurrence. We present a case of recurrent myoepithelioma in the nasal cavity with a palatal fistula treated both by a palatal approach and a piriform apertural approach. The combination of these approaches widens the surgical space, allowing removal of the mass. It is important not only to excise the mass but also to allow for reconstruction. (*Plast Reconstr Surg Glob Open* 2015;3:e508; doi: 10.1097/GOX.0000000000000491; Published online 8 September 2015.)

Myoepithelioma is a rare benign neoplasm arising in the major and minor salivary glands, which accounts for <1% of all salivary gland neoplasms.¹ Even less frequently, it occurs in the hard or soft palate. The recurrence rate has been reported to be 18%.² In this report, we present a case of recurrent myoepithelioma in the nasal cavity with palatal fistula diagnosed by a combination of palatal and piriform apertural approaches.

CASE REPORT

A 66-year-old woman, who underwent palatal tumor resection in another hospital 28 years ago, wore a palatal plate due to a remnant palatal fistula after surgery. She felt uncomfortable with her palatal plate, and her previous physician found that a tumor had grown through the palatal fistula. She was referred to otolaryngologists in our hospital and to us (plastic surgeons). The palatal fistula measured 10 mm from the oral side.

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Contrast-enhanced computed tomography showed a well-defined, heterogeneous, mildly enhancing soft-tissue mass arising from the soft palate (measuring 30 mm) (Fig. 1). The tumor was found attached to the inferior and middle nasal turbinate, but did not invade these structures. The bone defect of the palate was 20 mm in size.

The patient underwent incisional biopsy of the mass. Histological examination showed plasmacytoid cells; small polygonal cells with concentric or eccentric nuclei; dense, hyaline-abundant eosinophilic cytoplasm; and proliferative, abundant, and hematoxyphilic mucoid stroma. Ductal components were not observed. Mitotic activity was low, as indicated by an MIB-1 index of 6.04%.

Immunohistochemically, the biopsy specimen was positive for cytokeratin (AE1/AE3), smooth muscle actin (positive for focal area), glial fibrillary acidic protein, calponin, S-100, and p63, but was negative for epithelial membrane antigen and p53. Together, these findings were compatible with plasmacytoid-type myoepithelioma.

Surgery involved a trans piriform aperture approach by gingivobuccal incision and cleft palatal flap reconstruction of the palatal fistula. The palate was incised as in “2-flap palatoplasty,”³ which elevated the mucosa and periosteum off of the hard palate (Fig. 2). The mucosa around the fistula, which was confirmed as mucosal tissue, was elevated as a hinge flap to reconstruct the floor of the nasal cavity.

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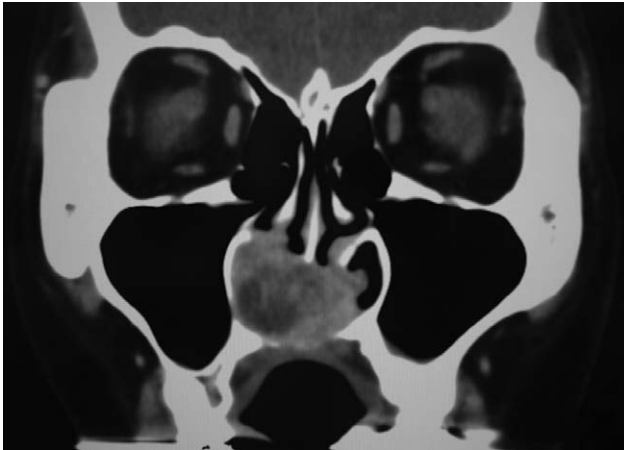


Fig. 1. CT scan reveals a recurrent tumor in the nasal cavity.

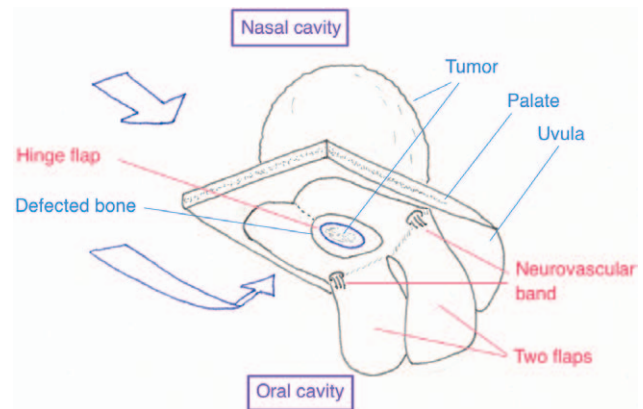


Fig. 3. Operation through oral and nasal cavities.

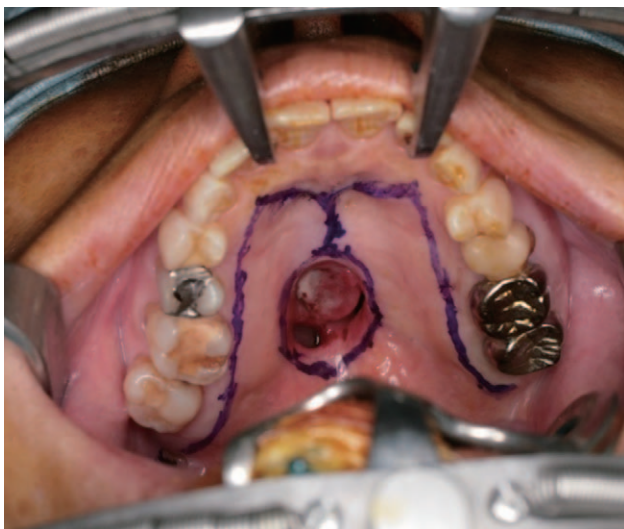


Fig. 2. Two palatal flaps.



Fig. 4. No recurrence has been observed after 6 months.

Simultaneously, we performed gingivobuccal incision. The mass was soft and bled easily and was easily detached from the bone. It was completely removed with submucosal dissection through a piriform aperture. The surgical specimen revealed a 27×20 mm well-circumscribed mass. We reconstructed the floor of the nasal cavity using the mucosa around the fistula and the palate by 2 flaps as in palatoplasty. The histological diagnosis was reconfirmed as myoepithelioma.

Postoperatively, the patient wore the palatal plate for only 1 month to protect the palate. There was no evidence of recurrence after 6 months (Figs. 3, 4).

DISCUSSION

We have presented a case of plasmacytoid-type myoepithelioma located in the nasal cavity. Myoepitheliomas usually present as slow-growing, painless, asymptomatic masses^{1,4,5} and are well-circumscribed,

solid tumors that usually measure <3 cm.^{4,5} In the present case, the mass grew slowly for 28 years with no pain. It was <3 cm, similar to cases reported in the literature.

Treatment of myoepithelioma is generally surgical excision with tumor-free margins according to tumor location.⁶ In the present case, the tumor located on the soft palate was removed completely with submucosal dissection.⁷

The recommended treatment is complete surgical excision.⁴ Endonasal endoscopic surgery^{8,9} and lateral rhinotomy^{10,11} have been previously reported. The present case represents the first report in which palatal and piriform apertural approaches with 2 palatal flaps as in palatoplasty were used to excise and remove the mass and to reconstruct the palatal fistula. Elevation of the palatal flaps during surgery allows the mass to be accessed. The mass was removed through a piriform aperture approach because it

was bigger than the bone defect of the palate. At the end of the surgery, we were able to perform palatoplasty to close the palatal fistula. Based on the size or features of a mass, Le Fort I osteotomy or piriform-extended osteotomy may need to be performed in such cases.

We adopted a palatal approach as done in 2-flap palatoplasty³ for 2 reasons: to widen the surgical space and to close the palatal fistula. Tongue flap or buccal musculomucosal flap procedures are not desirable for palatal fistula because myoepithelium is prone to recurrence and both types of flaps are pedicled flaps that require tissue from elsewhere in the mouth.

To the best of our knowledge, 3 cases in the literature have reported recurrence of myoepithelioma.^{11,12} Recurrence is correlated with positive margins at first excision.¹³ An overall recurrence rate of 18%² has been reported, and the recurrence rate is higher (42%) for those myoepithelial neoplasms with cytologically malignant features.² In the present case, the tumor most likely represents a recurrence rather than a new tumor due to the rareness of this tumor type (<1%¹) and its asymptomatic features.

Benign myoepithelioma can undergo malignant transformation, particularly in long-standing tumors or in tumors that frequently recur.⁵ However, the present case demonstrated low mitotic activity, as shown by an MIB-1 index of 6.04% histologically, and the nonadhesiveness of the mass to the bone and the lack of invasion clinically. Therefore, the tumor was classified as benign myoepithelioma despite being a long-term and recurrent case.

SUMMARY

Myoepithelioma is a rare, benign salivary neoplasm, most frequently located in the salivary gland; extrasalivary cases most commonly occur in the palate. This tumor is prone to recurrence. We present a case of recurrent myoepithelioma in the nasal cavity with a palatal fistula treated both by a palatal approach and a piriform apertural approach. The combination of these approaches widens the surgical space, allowing removal of the mass. It is important

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