

## Case Report

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# Clear Cell Myoepithelioma: A Rare Presentation in Nasal Cavity

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### ABSTRACT

Myoepitheliomas are predominantly tumors of salivary glands constituting less than 1% of all salivary gland tumors. A 65 year old women presented with bleeding unilateral mass for 03 months. Contrast Enhanced Computed Tomography (CECT) revealed a heterogeneously enhancing mass lesion arising from right anterior ethmoid air cells and filling the right nasal cavity entirely. Endoscope guided endoscopic biopsy was performed. Histopathological examination showed a well circumscribed tumour arranged in small glands and sheets. Tumor cell were immunopositive for S100 (Ib), Cytokeratin (CK) and Vimetin (focally ) while negative for chromogranin, synaptophysin, CD 10 and Smooth Muscle Actin (SMA). MIB-Labeling Index was <5%. With these features a diagnosis of clear cell myoepithelioma was made. Nasal myoepithelioma is an extremely rare low-grade neoplasm. Nasal myoepitheliomas are composed of myoepithelial cells with solid, myxoid or reticular patterns of growth. The cells themselves may be clear-cell type, spindle-shaped, plasmacytoid and epithelioid. In all reported cases of myoepithelioma, surgery was the mainstay treatment. Partial maxillectomy via a lateral rhinotomy approach, Caldwell-Luc procedure has been recommended for patients who were suspected to have a low-grade sarcomatous neoplasm. Only five cases have been reported in sinonasal region. We report the second case to be managed endoscopically. We report an extremely rare benign tumor of nasal cavity (clear cell myoepithelioma) which was managed endoscopically with recurrence within 6 months. The recurrence was also managed endoscopically. This case highlights the varied malignancies which may be encountered in sinonasal region.

**KEYWORDS:** Myoepithelioma; Clear cell tumor; Endoscopic sinus surgery.

### INTRODUCTION

Myoepitheliomas are predominantly tumors of salivary glands constituting less than 1% of all salivary gland tumors.<sup>1</sup> Only five cases of sinonasal myoepithelioma have been reported in literature till today's date. We present the first case of clear cell sub type of myoepithelioma in nose and the second case to be managed endoscopically.

### CASE REPORT

A 65 years old lady presented with complains of intermittent bleeding for 3 months and nasal obstruction on the right side for 2 months. There were no known comorbidities. On examination patient had a bulge over right lateral wall of nose seen externally. Anterior rhinoscopy revealed a fleshy smooth mass filling right nasal cavity. No blood clots or secretions were seen. Probing revealed the mass to be painless, soft in consistency, arising from right lateral wall of nose and bleeding on touch. Posterior rhinoscopy revealed normal nasopharynx. Contrast Enhanced Computed Tomography (CECT) revealed a heterogeneously enhancing mass lesion arising from right anterior ethmoid air cells and filling the right nasal cavity entirely (Figure 1). No features of bone erosion were seen. Hematoligical and biochemical parameters of patient were normal. It was planned an endoscope guided excision biopsy of mass. Intraopera-

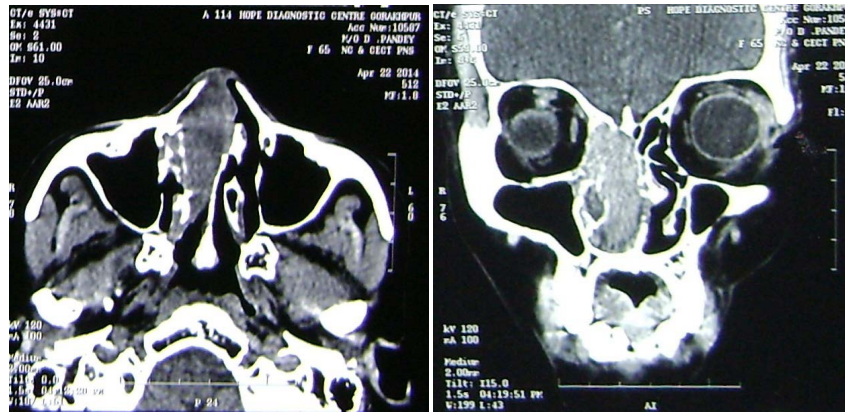


Figure 1: CECT Paranasal Sinuses: Heterogeneously enhancing soft tissue mass in right nasal cavity and anterior ethmoid air cells.

tive, mass was seen arising from sphenothmoidal recess. Brisk bleeding was encountered coming from sphenopalatine artery which was controlled with bipolar cautery. Post op uneventful.

Histopathological examination showed a well circumscribed tumour arranged in small glands and sheets. The individual cells were bland with clear cytoplasm. No layering was seen. Mitosis was scant and no necrosis was seen. Few dead bony spicules were noted (Figure 2). Special stains revealed glycogen in clear cells. Tumor cell were immunopositive for S100 (Ib), Cytokeratin (CK) and Vimetin (focally ) while negative for chromogranin, synaptophysin, CD 10 and Smooth Muscle Actin (SMA). MIB-1 Labelling Index was <5%. With these features a diagnosis of clear cell myoepithelioma was made.

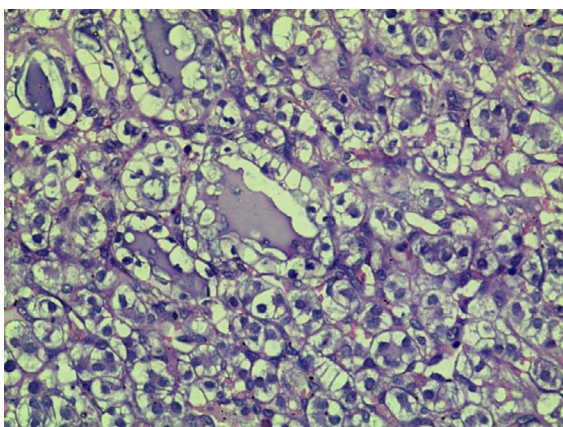


Figure 2: H&E microphotograph showing clear cell in tubules. No mitosis or atypia noted.

Patient was kept on follow up with no further episodes of epistaxis. Endoscopy after 6 months showed fleshy mass arising from middle meatus. Repeat tomography was done which showed recurrent mass lesion arising from right anterior ethmoid air cells. Endoscopic excision of middle turbinate and anterior ethmoid air cells was done and specimen sent for histopathology which also showed clear cell myoepithelioma with no evidence of malignant transformation. There has been no further recurrence during a follow up period of 1 year after second surgery.

DISCUSSION

Primary pleomorphic adenomas of the nasal cavity constitute around 18% of sinonasal nonepithelial neoplasms.<sup>2,3</sup> Nasal myoepithelioma is an extremely rare low-grade neoplasm. The main symptoms of nasal myoepithelioma are rapid enlargement of the tumor mass with nasal obstruction and epistaxis for periods varying from 3 months to 3 years.<sup>4,5</sup> The imaging appearance of a myoepithelioma is usually nonspecific.<sup>5</sup>

Nasal myoepitheliomas are composed of myoepithelial cells with solid, myxoid or reticular patterns of growth. The cells themselves may be clear-cell type, spindle-shaped, plasmacytoid and epithelioid.<sup>6</sup> Myoepitheliomas are usually devoid of ductal elements. However in three out of five cases of nasal myoepitheliomas small amounts of ductal elements have been reported. Variable degree of nuclear atypia, often mixed with a population of cells with eosinophilic cytoplasm has been noted. Frankly malignant change has not been seen in nasal myoepitheliomas. Immunohistochemistry (IHC) is an important adjunct in differential diagnosis of myoepitheliomas (Figures 3 and 4). The possible differential diagnosis and their features are as in Table 1.<sup>7</sup>

In all reported cases of myoepithelioma, surgery was the mainstay treatment. Partial maxillectomy via a lateral rhinotomy approach, Caldwell-Luc procedure has been recommended for patients who were suspected to have a low-grade sarcomatous neoplasm.<sup>4,8</sup> With the advent of nasal endoscopes Fujukura and Okubu removed a 10 mm tumor endoscopically.<sup>9</sup> In our patient, CECT showed tumor arising from anterior ethmoid air cells, restricted to nasal cavity with no bony erosion. Hence, an endonasal endoscopic approach was planned.

CONCLUSION

We report an extremely rare benign tumor of nasal cavity (clear cell myoepithelioma) which was managed endoscopically with recurrence within 6 months. The recurrence was also managed endoscopically. This case highlights the varied malignancies

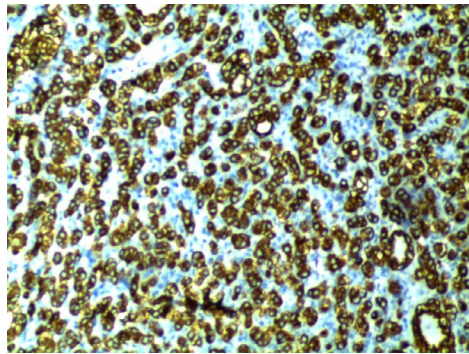


Figure 3: IHC showing membranous cytokeratin positivity.

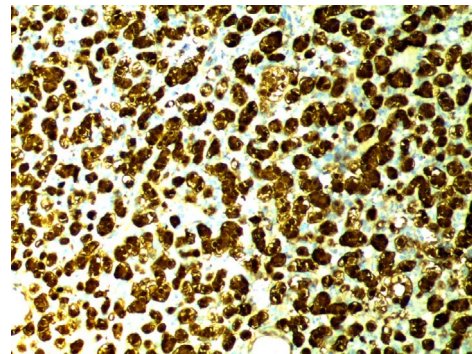


Figure 4: IHC showing diffuse, strong nuclear positivity for S100.

S.No.	Tumor	Morphology	IHC & Special stain
1	Clear cell oncocytoma	Trabecular pattern in cell with central round nuclei and granular eosinophilic cytoplasm	PTAH + Vimentin +/-
2	Epithelial myoepithelial Carcinoma	Bilayer ductal structure, inner cuboidal & outer clear cell	Inner layer – CK + Outer cells – S 100 +, Actin +
3	Mucopidermoid Carcinoma	Squamoid and intermediate cell along with clear cells	Clear cell resistant to diastase, Mucicarmine +
4	Acini cell carcinoma	Microcystic pattern cells with peripheral nuclei and basophilic cytoplasm	PAS + (not sensitive to diastase), Amylase +, CEA +
5	Clear cell carcinoma NOS	Usually diagnosis of exclusion	Negative myoepithelial marker (SMA, S100, Calponin, P63 – ve)
6	Metastatic RCC	Sinusoidal pattern clear cells, Hemosiderin deposits, Haemorrhage present.	CK +, Vimentin +, CD 10 +

Table 1: Histopathological and IHC characteristics of differential diagnosis of myoepitheliomas.

which may be encountered in sinonasal region. An in-depth knowledge, high index of suspicion and use of IHC is required for an accurate diagnosis. Endoscopic excision is likely to be the mainstay of management in the future.

**CONFLICTS OF INTEREST:** None.

**CONSENT**

Authors obtain written informed consent from the patient for submission of this manuscript for publication.

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