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Esophageal Myoepithelial Carcinoma - Single Case Report

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Abstract: ***Myoepithelial carcinoma (MC) is a rare malignant neoplasm that mainly occurs in the salivary gland. (1) It was first reported by Stromeyer et al (2) in 1975. MC has been identified in several locations, including the parotid gland, minor salivary glands (especially the palate), respiratory tract, maxillary sinus, larynx, lungs, and penile. The most common site is the parotid gland. Patients often do not exhibit obvious symptoms in the early stage of an oesophageal tumour. As the tumour grows, patients present with different symptoms that are dependent on tumour size and location, including progressive dysphagia, chest pain, and hoarseness. Since the tumor is extremely rare and there is a lack of knowledge of aggressiveness of the condition, available treatment is uncertain. According to previous reports, surgery remains the best treatment option for MC at other sites. Here, we report a case of MC of the oesophagus in a 47-year-old male that was treated with trans hiatal esophagectomy.***

Keywords: Myoepithelial carcinoma, rare, malignant, neoplasm, salivary gland

1. Case Summary

A 47 years old male presented with complaints of dysphagia for 6 months more for solids than liquids associated with epigastric pain and with history of loss weight of around 12kgs in 3 months, with no history of Malena and jaundice. He has history of pulmonary tuberculosis in 2004, and completed ATT for the same . No other comorbidities.

Ugi scopy was suggestive of ulceroproliferative growth 28-32cms from incisors involving mid esophagus, biopsy suggestive of poorly differentiated squamous cell carcinoma.

PET-CT was suggestive of mid esophageal growth involving subcarinal and retro cardia esophagus for a length of 5.2cm with no surrounding organ involvement and no distant metastasis.

Patient underwent Trans hiatal esophagectomy with feeding jejunostomy, postoperatively Patient recovered without any complications.

Postoperatively histopathology was reported as : Pt3, Pn0, High grade myoepithelial carcinoma. 9 nodes were examined and none of them were involved.

2. Histopathology

Figure 1: Gross image showing the esophageal mucosa appearing unremarkable. Tumor bulk is noted in the submucosal region

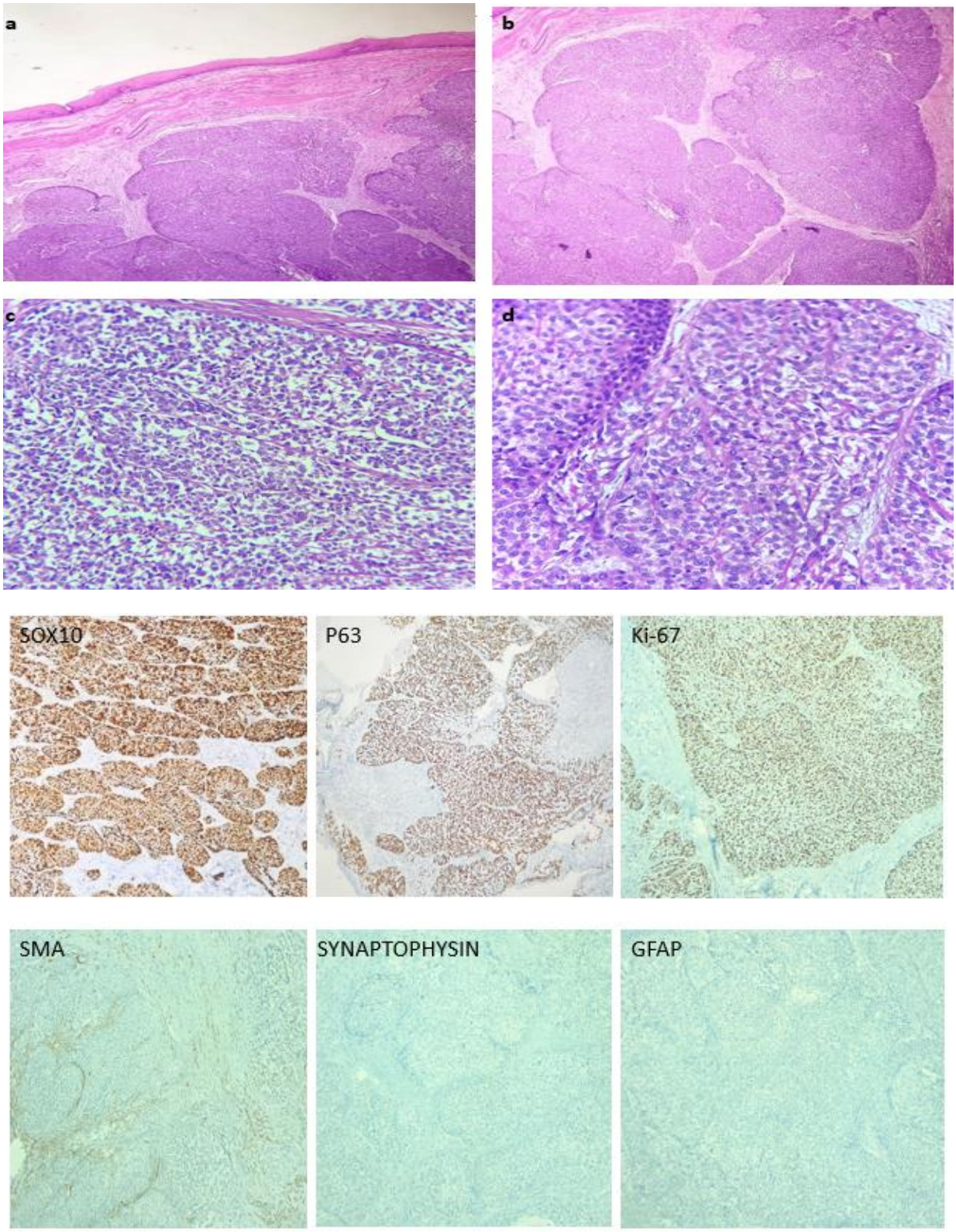


Figure 2: Light Microscopy

a) H &E image showing squamous epithelium with the subepithelium showing tumour arranged in nests and sheets X 40
b) At low power the tumour cells show a nodular growth pattern with hyalinized stoma
c) Tumour cells have scant to cytoplasm with monomorphic nuclei and low mitotic rate X400
d) Foci of myoepithelial carcinoma with clear cell features highlighted by vacolated empty looking clear cytoplasm X400

Radiology

- FDG AVID MASS LESION (LENGTH – 52 mm; SUV max: 19.91) INVOLVING THE SUBCARINAL AND RETROCARDIAC ESOPHAGUS -- CARCINOMA ESOPHAGUS.
- NO LYMPH NODE / DISTANT METASTASIS.

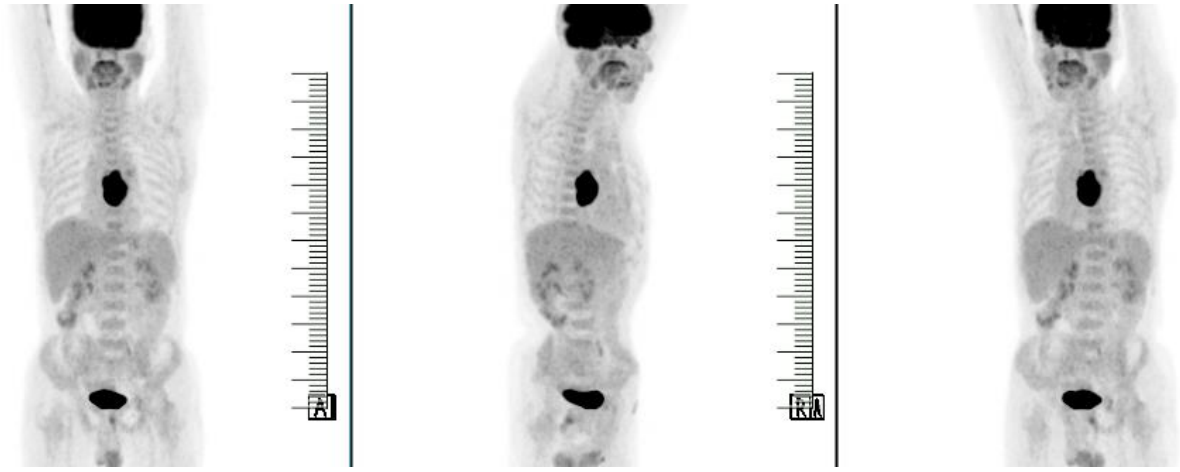
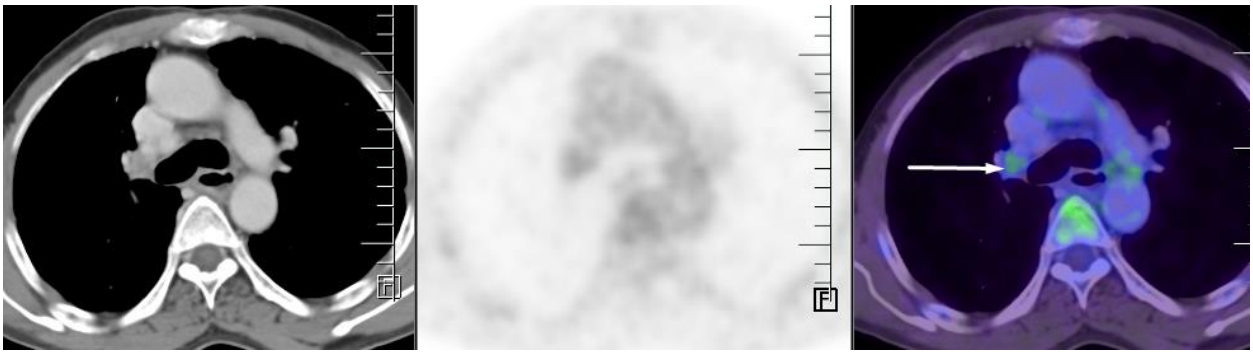


Figure 3: Immunohistochemistry Panel for SOX 10, P63 showing strong and diffuse positivity. Ki 67 Labelling index is 80% and negative for SMA, Synaptophysin and GFAP

Enlarged lymph nodes in

- **RIGHT HILUM (13 X 11 MM; SUV MAX: 2.10)**
- **LEFT HILUM (6 X 4 MM; SUV MAX: 3.02)**

--- *Reactive*



3. Discussion

The combination of histopathology and immunohistochemistry has diagnostic significance for myoepithelial carcinoma[3]. Specifically, myoepithelial differentiation and tumor infiltration into adjacent tissues are the currently accepted diagnostic criteria[4]. MC is a rare biphasic tumor with low malignant potential that accounts for 1% of salivary gland tumors . The most common location is parotid, and a small proportion occurs in the submandibular gland and small salivary gland . In addition to the salivary glands, MC rarely occurs in other parts of the body, oral cavity, or neck[5]. MC can also originate in the chest, lungs, skin, and stomach[6-8]

The definitive diagnosis of MC depends on the results of pathological examination and immunohistochemistry. Myoepithelial carcinomas (malignant myoepitheliomas) are unencapsulated and usually multinodular in appearance, with infiltrative borders. The epithelioid cells are large polygonal cells with central ovoid or round nuclei and eosinophilic or amphophilic, sometimes focally clear cytoplasm. Myoepithelial carcinoma (malignant myoepithelioma) mainly exhibits different types of tumor matrix: myxoid and hyalinized. Myoepithelial cells are seen.

The following markers are found to be useful immunohistochemical markers in myoepithelial carcinoma (malignant myoepithelioma): Cytokeratins (eg, AE1/AE3, CK 5/6, Cam 5.2, CK7, CK14), Vimentin (reported to be positive in neoplastic myoepithelial cells and negative in normal myoepithelial cells), S-100 (almost always positive in the neoplastic myoepithelial cells but not in the normal myoepithelial cells), SOX-10, Ki 67 and p63. [9]

Due to the rarity and unproven malignant potential of these tumors, the definite treatment protocol is still unknown. According to previous reports, surgery remains the safe and effective treatment option for MC of other sites in the body. Complete surgical excision of the tumor provides superior outcomes in terms of survival and recurrence rates. Due to the lack of sufficient reported cases, there is insufficient evidence for postoperative chemoradiotherapy. In our case, we performed surgical treatment based on our experience with oesophageal cancer. At 4 months of follow-up, the patient was asymptomatic.

4. Conclusion

In conclusion, the present study described a rare case of oesophageal MC in a male patient. To the best of our knowledge, a case of MC in the oesophagus has been reported

in less than 10 cases worldwide. Identifying the diagnosis and effective treatment remains a challenge. However, complete surgical excision is still considered the most effective treatment.

Conflict of interest statement

Conflict-of-interest statement: The authors declare that they have no conflict of interest.

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