Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology 26 (2014) 580–584

Contents lists available at ScienceDirect

Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology

journal homepage: www.elsevier.com/locate/jomsmp

Case report

# Soft tissue myoepithelial carcinoma of neck: A rare case report with review of literature

Niharika Swain*, Shwetha V. Kumar, Jigna Pathak, Shilpa Patel

*Department of Oral Pathology, MGM Dental College and Hospital, Junction of NH-4 and Sion Panvel Expressway, Sector-18, Kamothe, Navi Mumbai 410 209, Maharashtra, India*

ARTICLE INFO

*Article history:*
Received 18 March 2013
Received in revised form 21 June 2013
Accepted 11 July 2013
Available online 17 September 2013

*Keywords:*
Soft tissue tumor
Myoepithelial neoplasms
p63
Imunoexpression

ABSTRACT

Primary soft tissue myoepithelial tumor, an uncommon variant of myoepithelial neoplasms, has been recently described in reviewed literature. We report a rare case of soft tissue myoepithelial carcinoma in a 60 year old male patient presented as a unilateral neck mass. MRI showed a large lobulated infiltrating heterointense mass with central necrotic area involving left parapharyngeal space. Histopathological examination revealed multilobular growth pattern of epitheloid cells with marked atypia and frequent mitosis. Immunohistochemistry was reactive for p63, calponin, CD10, pancytokeratin, EMA and podoplanin with high (40%) proliferative labeling index. Except for the presence of increased malignant cell population, soft tissue myoepithelial carcinoma mimics their salivary gland counterpart in clinical and biologic behavior. Rarity in head and neck region (only 16%) and heterogeneity in cellular morphology and architectural patterns, may lead to misdiagnosis of extraglandular myoepithelial carcinoma. So, careful and meticulous observation of both histopathologic and immunophenotypic features are essential for correct diagnosis of such entities.



## 1. Introduction

Myoepithelial neoplasms, initially recognized to be of salivary gland origin, in recent years are known to arise from other primary sites like skin, nasal cavity, paranasal sinuses, breast, lacrimal glands, bronchus, lungs or kidneys [1]. In 1995, the first case of soft tissue myoepithelioma in retroperitonium was reported by Burke et al. Two years later, a case series of 19 cases of soft tissue myoepitheliomas was published by Kilpatrick et al. [2]. This uncommon soft tissue counterpart was included as a separate entity in World Health Organization Classification of soft tissue tumors in 2002 [3]. In 2003, Hornick et al. published the largest case series of 101 cases of this entity with proposal of criteria for malignancy and prognostic parameters [4]. In 2007, 29 cases of soft tissue myoepithelial carcinoma in pediatric population were reported by Gleason et al. The histogenesis of these extraglandular neoplasms is poorly understood [5]. Due to wide variation in morphologic and immunophenotypic expression of myoepithelial cells, it is challenging to distinguish these neoplasms from other soft tissue tumors, when arising in unusual sites. Furthermore, as these neoplasms are extremely rare in head and neck region, soft tissue myoepithelial tumors are more liable to misdiagnosis.

## 2. Case report

A 60-year-old male patient reported with a large rapidly growing unilateral neck swelling since 2 months (Fig. 1 A). The swelling was non-tender with no associated symptoms. The magnetic resonance imaging (MRI) showed a large lobulated infiltrating heterointense mass extending medially anterior to the carotid space in left parapharyngeal space, posteriorly into the posterior triangle of neck, inferiorly along the lateral aspect of carotid space in submandibular region and superiorly upto the parotid space. The lesion did not show any parotid gland involvement. On post contrast evaluation there was moderate heterogenous enhancement of the lesion with large non-enhancing well defined necrotic area in the center (Fig. 1B and C). An incisional biopsy revealed poorly differentiated carcinoma. The patient further underwent wide surgical tumor resection including radical neck dissection. Intra-operatively, no continuity with regional major salivary glands was observed (Fig. 2 A and B).

Histopathologically, multilobular growth pattern with nests of predominantly epitheloid cells with areas of necrosis and hemorrhage was noted. In some areas spindle and clear cell

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* Corresponding author. Tel.: +91 93 24701036; fax: +91 22 27423185.
*E-mail address:* niharikadec30@gmail.com (N. Swain).

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http://dx.doi.org/10.1016/j.ajoms.2013.07.014

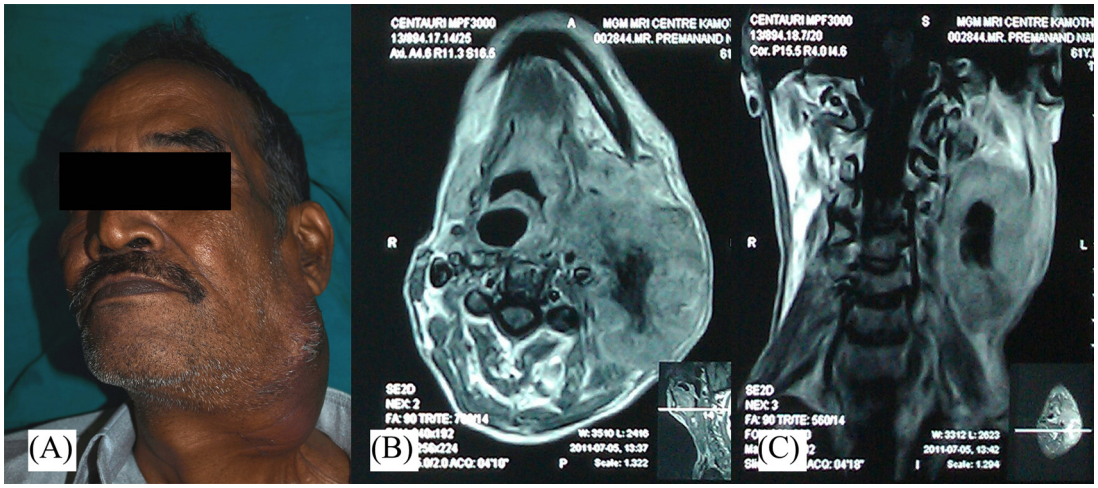


**Fig. 1.** (A) Preoperative photograph of the patient showing a swelling in the neck (left side). (B and C) Preoperative magnetic resonance imaging scan showing heterointense mass with central nonenhanced necrotic area.

morphology with collagenous spherules were observed. Some tumor islands showed squamous differentiation. The epitheloid cells with moderate to abundant eosinophilic cytoplasm showed nuclear pleomorphism, vesicular or coarse chromatin, and prominent nucleoli. Within the neoplastic nodules, vacuolated cells with signet ring like appearance were also frequently encountered. The mitotic count was high, showing more than 25 mitoses per 10 high power fields (HPF) (Fig. 3 A–D). Immunochemistry revealed intense staining for p63 (Prediluted, Ready to use, clone 4B1E12, Biogenex, USA), calponin (Prediluted, Ready to use, clone CALP, Biogenex, USA) and CD10 (Prediluted, Ready to use, clone 56C6, Biogenex, USA). Ki67 (Prediluted, Ready to use, clone BGX-Ki-67, Biogenex, USA) immunostaining revealed a tumor proliferative labeling index of 40%. Immunostaining showed intense positivity for, multifocal positivity for epithelial membrane antigen (Prediluted, Ready to use, clone E29, Biogenex, USA), pancytokeratin (Prediluted, Ready to use, clone AE1/AE3, Biogenex, USA) and podoplanin (Prediluted, Ready to use, clone D2-40, Thermo Scientific, USA) (Fig. 4 A–G).

The staining was negative for Smooth Muscle Actin SMA (Prediluted, Ready to use, clone 1A4, Biogenex, USA), GFAP (Prediluted, Ready to use, clone GA-5, Biogenex, USA) (Fig. 4H and I), S100 (1/2000, rabbit polyclonal, Dako, Glostrup, Denmark), cytokeratin 7 (Prediluted, Ready to use, clone OV-TL12/30, Biogenex, USA) and 10 (Prediluted, Ready to use, clone DEK-10, Biogenex, USA). The radical neck dissection specimen showed invasion in three lymph nodes with extracapsular spread. The histopathological appearance and immunophenotype indicated very well with a myoepithelial carcinoma, apparently arising in the soft tissue. Differential diagnosis of soft tissue myoepithelial carcinoma has broad range due to the cytological and immunophenotypic heterogeneity of myoepithelial cells which includes high grade extra skeletal myxoid chondrosarcoma, epitheloid malignant peripheral nerve sheath tumor, poorly differentiated synovial sarcoma, epitheloid sarcoma, sclerosing epitheloid fibrosarcoma, metastatic carcinoma, and metastatic melanoma. Careful observation of histopathologic features and myogenic immune expression is crucial for distinguishing from aforementioned neoplasms.

Following surgical resection, the patient was given adjuvant radiotherapy. The patient however succumbed to the tumor within 6 months of initial diagnosis.

## 3. Discussion

Extraglandular myoepithelial tumor shows a high predilection toward the limbs and limb girdles. In the head and neck region they account for only 16% of all reported soft tissue myoepitheliomas

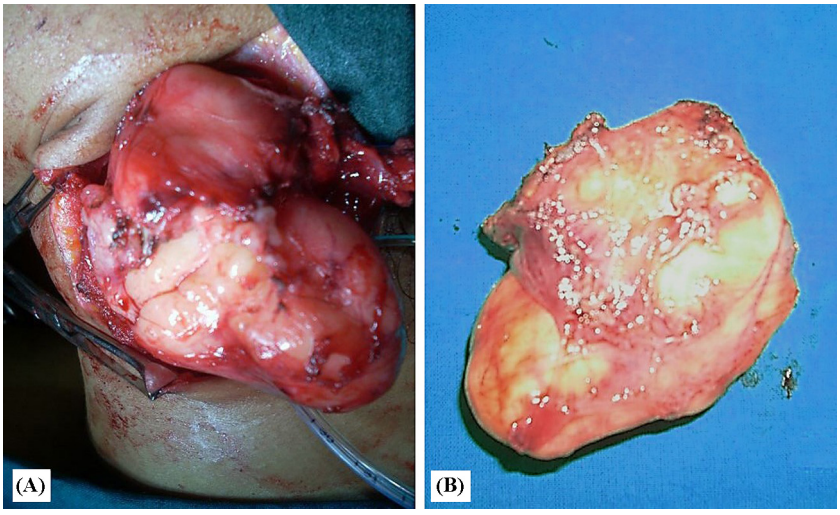


**Fig. 2.** (A) Intraperative view of tumor mass. (B) Macroscopic view after resection showing a hard, nodular tumor.