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Oncocytic Mucoepidermoid Carcinoma: A Case Report

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Abstract

Oncocytes are large epithelial cells characterized by abundant eosinophilic granular cytoplasm containing excessive mitochondria. Mucoepidermoid Carcinoma (MEC) is the most common salivary gland malignancy (10% of all major gland tumors), most commonly found in the parotid gland. Oncocytic Mucoepidermoid Carcinoma (OMEC) is a very rare tumor found most commonly in the parotid gland. This was a case of oncocytic mucoepidermoid carcinoma in a 26 year old Vietnamese male. He initially presented with a painless mass over his left jaw. Biopsy pathology of the mass showed lesion cells with no high-grade features identified; complete excision was recommended for further classification. Microscopic examination showed an ill-defined infiltrative tumor composed predominantly of solid sheets and nests. The tumor cells were predominantly oncocytic characterized by abundant eosinophilic cytoplasm, round nuclei, and prominent nucleoli with focal pleomorphism. Scattered mucocytes were present. To aid in diagnosis, the following stains were done: CK5/6, CK7, CK14, p63, Ki-67, CK903, CK20, GCDFP, SOX10, Androgen receptor, GATA3, PAS-D, and mucicarmine. The stains CK5/6 and p63 were performed to identify oncocytes. PAS-D and mucicarmine were performed to identify mucin and mucous cells. An accurate diagnosis of OMEC can be made using a combination of histological evaluation and the appropriate immunostains and special stains. Equivocal staining results can be further evaluated for MAML2 translocation with RT-PCR or FISH.

Introduction

Oncocytes are large epithelial cells characterized by abundant eosinophilic granular cytoplasm containing excessive mitochondria [1-3]. Histochemical studies have shown similarities between oncocytes and intercalated duct reserve cells in oxidative enzyme concentrations [3]. Oncocytes can be found in the salivary glands, kidney, thyroid, parathyroid, pituitary, adrenal glands, liver, pancreas, fallopian tubes, testes, stomach, and bronchi [2,3]. These cells may occur as metaplasia or age related degeneration, or may occur as a lesion in neoplastic or hyperplastic processes [1-3]. Within the salivary glands, oncocytic lesions can be classified into four groups:

1. Malignant oncocytic tumors,
2. Oncocytic variants of other salivary gland tumors,
3. Oncocytic hyperplasia,
4. Oncocytomas [1].

Oncocytic variants have been reported in almost all salivary gland tumors, including: mucoepidermoid carcinoma, acinic cell carcinoma, adenoid cystic carcinoma, adenocarcinoma, lipoadenoma, salivary duct carcinoma, and myoepithelioma [1,3-6].

Mucoepidermoid Carcinoma (MEC) is the most common salivary gland malignancy (10% of all major gland tumors), most commonly found in the parotid gland [3-5,7-9]. MEC arises from salivary gland ducts and is characterized by mucous cells, intermediate cells, and epidermoid (squamoid) cells [2,3,5,7-9]. Low grade lesions are characterized by cystic spaces with many mucous cells, while high grade lesions are characterized by solid areas of intermediate and epidermoid cells with necrosis and neural involvement [3]. There are many different variants of MEC, including: sebaceous, clear, spindle, melanocytic, goblet, sclerosing, psammomatous, unicystic, and oncocytic [3,5,9].

Oncocytic Mucoepidermoid Carcinoma (OMEC) is a very rare tumor (less than 40 reported cases in the literature) that has been reported in salivary glands (most often parotid) as well as in lacrimal gland, bronchus, palate, neck, and trachea [1,4-9]. This tumor is characterized by predominantly oncocytic cells, epidermoid cells, and mucous goblet cells [3,4,6]. While there is

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Table 1: Staining to aid in the diagnosis of OMEC.

Stain	Finding
PTAH	(+) in oncocytes; (-) in acinic cell carcinoma
Alcian blue	(+) in mucous cells
Mucicarmine	(+) in mucous cells
PAS-D	(+) in mucous cells
Anti-mitochondrial ab-1	(+) in oncocytes
Anti-S-100	(-) in oncocytes
Anti-vimentin	(-) in oncocytes
Anti-Smooth muscle actin	(-) in oncocytes
p63	(+) in oncocytes; nuclear positivity in OMEC, peripheral positivity in oncocytoma, oncocytic carcinoma (-) in acinic cell carcinoma, salivary duct carcinoma
CK 5/6	(+) in oncocytes
AE1/AE3	(+) in mucous cells
Androgen receptor	(+) in salivary duct carcinoma (-) in OMEC
GATA3	(+) diffuse staining in salivary duct carcinoma; (+) positive but not diffuse staining in other salivary gland tumors
SOX10	(+) in acinic cell, adenoid cystic, epithelial-myoepithelial, myoepithelial carcinomas and pleomorphic adenoma
GCDFP	(+) in salivary gland carcinoma
CK20	(+) in small cell carcinoma of major salivary glands
CK903	(+) in myoepithelial carcinoma
CK14	(+) in salivary gland tumors except acinic cell carcinoma
CK7	(+) in salivary gland carcinoma

no well-defined proportion of oncocytes for making a diagnosis of OMEC *vs.* MEC with oncocytic metaplasia, a review of previously reported cases finds that almost all authors describe >50% oncocytes when making a diagnosis of OMEC [3,6,9]. OMEC usually presents as a low or intermediate grade tumor with a good prognosis [5,8,9]. It is unknown if oncocytic differentiation has any effect on prognosis [1]. The age range of reported OMECs is 20 to 80 years, with a slight male predominance [5]. The differential diagnosis of OMEC includes: oncocytosis, oncocytoma, oncocytic carcinoma, Warthin's tumor, pleomorphic adenoma with oncocytic change, and oncocytic lipoadenoma [4].

The diagnosis of OMEC can be challenging due to overlapping histological features; distinguishing this tumor from an oncocytoma, oncocytic carcinoma, or other oncocytic neoplasms requires adequate sampling, detailed histologic study, immunohistochemical stains, special stains, and possibly Florescence *In Situ* Hybridization (FISH) testing or Reverse Transcriptase-Polymerase Chain Reaction (RT-PCR) [3,6,9]. Transmission electron microscopy and immunoelectron microscopy have also been used [1]. The most important histological feature of OMEC may be the presence of mucous cells; mucous gland cells can rule out oncocytosis, oncocytoma, and oncocytic carcinoma [4,7]. Stains that have been used to help diagnose OMEC include: Phosphotungstic Acid Hematoxylin (PTAH), Alcian blue, antibody to mitochondria AB-1, anti-S-100, anti-vimentin, anti-smooth muscle actin, p63, CK5/6, AE1/AE3, androgen receptor (details listed in (Table 1). Staining to aid in the diagnosis of OMEC) [1,3-6,9,10]. The detection of MECT1-MAML2 or CRTC1-MAML [2] fusion transcript can be identified by RT-PCR or FISH; the presence of a MAML2 translocation can help identify the MEC component of OMEC [3,6,9]. The accurate diagnosis of OMEC is important for patient management [1].

This case report aims to raise awareness of the very rare tumor OMEC and the different stains that can be used to aid in its diagnosis.

Case Presentation

This was a case of oncocytic mucoepidermoid carcinoma in a 26 year old Vietnamese male. The timeline of the patient management is detailed in (Table 2). Timeline of Events, He initially presented with a painless mass over his left jaw that he first noticed 9 months ago. He denied pain, intermittent swelling, foul taste, paresthesia, facial nerve weakness and redness to the area, exposure to pet cats, travel outside of the country, fever, chills, cough, weight loss, dysphagia, or dyspnea. He was recently diagnosed with essential hypertension. He denied any prior surgeries. He reported no known allergies. He denied any history of cancer in his family. He denied tobacco and alcohol use. His only medication was carvedilol. Review of systems was unremarkable.

On physical exam there was facial asymmetry with left sided facial swelling. There was no erythema. Palpation over the left parotid gland revealed a 3 cm somewhat well-defined mass. There was clear salivary flow from both Stensen's ducts. The remaining physical exam was unremarkable. A CT soft tissue neck with contrast showed a rounded somewhat ill-defined in homogeneously enhancing mass involving the anterior aspect of the left parotid gland and posterior aspect of the adjacent masseter muscle, measuring 3.9 cm × 3.1 cm × 3.6 cm. Multiple nonspecific cervical lymph nodes were also evident including inferior left periparotid nodes, left submandibular nodes, and internal jugular nodes more on the left, all of which demonstrated homogenous density and well-defined margins. An ultrasound guided aspirate biopsy (multiple with 18 gauge needle) of the left parotid mass was performed; the imaging showed an enlarged hyperemic left parotid gland demonstrating multiple solid and single 6 mm × 6 mm cystic masses. The biopsy pathology showed lesion cells with no high-grade features identified; complete excision was recommended for further classification.

Table 2: Timeline of Events.

Date	Event	Outcome
1/14/2019	Presented to ED for evaluation of painless left jaw mass for past 9 months	Imaging ordered
1/14/2019	CT Soft Tissue Neck with Contrast	3.9 cm × 3.1 cm × 3.6 cm ill-defined in homogeneously enhancing mass along anterior aspect left parotid gland and posterior aspect of adjacent masseter muscle
1/20/2019	Initial consult with oral & maxillo-facial surgery	Recommended US guided biopsy of parotid mass
1/30/2019	US guided aspirate biopsy left parotid mass	Pathology: lesional cells present with no high-grade features identified – recommend complete excision
2/13/2019	Seen by general surgery clinic	Complete excision recommended
03-01-19	Left subtotal parotidectomy	Intraoperative frozen section was not diagnostic for malignancy.

Table 3: Immunohistochemical Stains & Special Stains.

Immunohistochemical/Special Stain	Result
CK5/6	Positive
CK7	Positive
CK14	Weak positive
p63	Diffuse nuclear positivity
Ki-67	<10%
CK903	Positive
CK20	negative
GCDFP	Negative
SOX10	Negative
Androgen receptor	Interpreted as negative
GATA3	Interpreted as negative
PAS-D	Positive intracytoplasmic globules
Mucicarmine	Intracellular positivity in mucocytes

The decision was made to proceed with a left subtotal parotidectomy to achieve complete excision. Subcutaneous flaps were made over the parotid capsule and platysma, and the superficial parotid gland was bluntly dissected out. There was a firm, suspicious mass present in the gland which involved the masseter; deeper parotid tissue, en bloc with part of the masseter muscle, was excised in order to remove the suspicious tissue. The facial nerve was identified and preserved, with the exception of the buccal branch, which coursed straight into the lesion. Intraoperative frozen section was not diagnostic for malignancy; neck dissection was not performed.

Materials and Methods

Grossly the specimen was a left parotid gland weighing 65 g and measuring 10 cm × 7 cm × 2 cm. Sectioning revealed a tan firm lobular mass measuring 4 cm × 3 cm × 3 cm that appeared to be partially encapsulated and surrounded by normal appearing parotid parenchyma. Representative sections were sampled and submitted in 17 cassettes, including frozen sections and normal uninvolved parotid gland. Two left parotid lymph nodes were also resected; grossly these appeared as irregularly shaped fragments of tan-brown tissue measuring 1 cm × 1 cm × 0.5 cm and 1 cm × 0.3 cm × 0.2 cm; each fragment was bisected and submitted in a single cassette.

Results and Discussion

Microscopic examination showed an ill-defined infiltrative tumor composed predominantly of solid sheets and nests. Focal (<20%) areas of the tumor showed cystic and microcystic areas. The tumor cells were predominantly oncocytic characterized by abundant eosinophilic cytoplasm, round nuclei, and prominent nucleoli with focal pleomorphism. Scattered monocytes were present. Mitotic figures were not identified. Intermixed and peripheral chronic inflammations were present; Sialadenitis was present. Focal lymphovascular invasion and perineural invasion were present. Adjacent to the tumor was uninvolved parotid gland tissue and uninvolved fibro connective tissue including skeletal muscle. Results of immunohistochemical stains and special stains performed are detailed in (Table 3). Immunohistochemical Stains and Special stains. One surgical margin was involved by carcinoma. No metastatic carcinoma was identified in two lymph nodes (0/2). The overall findings supported the diagnosis of mucoepidermoid carcinoma, intermediate grade (modified Healy and Memorial Sloan Kettering Cancer Center's grading schemes used), 4 cm, oncocytic variant; pathologic TMS staging was pT2N0.

Based on the pathologic diagnosis of intermediate grade mucoepidermoid carcinoma, oncocytic variant, pT2N0, the patient will receive cervical neck dissection and radiation therapy, and likely chemotherapy. A rare case of oncocytic mucoepidermoid carcinoma in a 26 year old male was presented. To aid in this diagnosis, the following stains were done: CK5/6, CK7, CK14, p63, Ki-67, CK903, CK20, GCDFP, SOX10, Androgen receptor, GATA3, PAS-D, mucicarmine. The stains CK5/6 and p63 were performed to identify oncocytes. PAS-D and mucicarmine were performed to identify mucin and mucous cells. Androgen receptor and GATA3 were performed to rule out salivary duct carcinoma. CK7, CK14, and GCDFP were performed to rule out salivary gland carcinoma. CK20 was performed to rule out small cell carcinoma. CK903 was performed to rule out myoepithelial carcinoma. SOX-10 was performed to rule out acinic cell, adenoid cystic, epithelial-myoepithelial, myoepithelial carcinomas and pleomorphic adenoma. Ki-67 was performed to evaluate mitotic activity. Based on the histological evaluation and results of the immunostains and special stains, the diagnosis of OMEC was made. In this case it was not necessary to order RT-PCR or FISH to test for MAML2 translocation. However, if the results of staining were equivocal MAML2 testing would have been the next step.

Conclusion

This case report provides a comprehensive list of stains that are useful in the diagnosis of OMEC. The main limitation of this report is that all stains presented were not used in this case; therefore they were not all compared for usefulness in making the diagnosis (stains were chosen based on pathologist preference). The main lessons of this case report are:

1. OMEC is a rare tumor found most commonly in the parotid gland and

2. Although diagnosis can be challenging, the use of proper histological evaluation, immunostains, and special stains can lead to an accurate diagnosis of OMEC.

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References

1. Brannon RB, Willard CC. Oncocytic mucoepidermoid carcinoma of parotid gland origin. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2003;96(6):727-73.
2. Akhtar K, Qadri S, Ray PS, Sherwani RK. Oncocytoma of the parotid gland: Dilemma for the cytopathologist. J Orofac Sci. 2016;8(1):66-70.
3. Kumar R, Natarajan S, Sneha KS, Chitra NS, Boaz K, Manaktala N. Oncocytes in Mucoepidermoid Carcinoma of the Palate: Diagnostic Challenges. Case Rep Dent. 2017;2017:5741821.
4. Deveci MS, Deveci G, Gunhan O, Finci R. Oncocytic mucoepidermoid carcinoma of the parotid gland: Report of a case with DNA ploidy analysis and review of the literature. Pathol Int. 2000;50(11):905-9.
5. Kwon H, Lim W, Choi Y, Nam J, Han C, Kim J, et al. High-grade oncocytic mucoepidermoid carcinoma of the minor salivary gland origin: a case report with immunohistochemical study. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2010;109(6):e72-7.
6. Fujimaki M, Fukumura Y, Saito T, Mitani K, Uchida S, Yokoyama J, et al. Oncocytic mucoepidermoid carcinoma of the parotid gland with CRTC1-MAML2 fusion transcript: report of a case with review of literature. Hum Pathol. 2011;42(12):2052-5.
7. Wade TV, LiVolsi VA, Montone KT, Baloch ZW. A Cytohistologic Correlation of Mucoepidermoid Carcinoma: Emphasizing the Rare Oncocytic Variant. Pathol Res Int. 2011;(3):135796.
8. D'Antonio A, Boscaino A, Caleo A, Addesso M, Orabona P. Oncocytic variant of mucoepidermoid carcinoma: A diagnostic challenge for the pathologist. Indian J Path Microbio. 2015;58(2):201-3.
9. Liao X, Haghighi P, Coffey CS, Xu X. A rare case of exclusively oncocytic mucoepidermoid carcinoma with MAML2 translocation. Rare Tumors. 2016;8(2):6166.
10. Pathologyoutlines.com. Michigan: Pathology Outlines; c2019.