ORIGINAL RESEARCH PAPER | Radiology | Volume : 6 | Issue : 11 | November 2016 | ISSN - 2249-555X | IF : 3.919 | IC Value : 74.50

Mucoepidermoid Carcinoma of Maxillary Sinus : A Case Report and Review of Literature

KEYWORDS: Mucoepidermoid carcinoma , maxillary sinus, salivary gland malignancies.

Yumkhaibam Sobita Devi	Shoibam Subhaschandra Singh	Dulasiraman Ponnas
Assistant Professor, Department of Radiotherapy, Regional Institute of Medical Sciences, Imphal, Manipur, 795004-India	Associate Professor, Department of Radiology, Regional Institute of Medical Sciences, Imphal, Manipur, 795004-India	Postgraduate Student, Department of Radiotherapy, Regional Institute of Medical Sciences, Imphal, Manipur, 795004-India

Himadri Kumar Daru	Laishram Jaichand Singh	R.K. Tamphasana Devi
Postgraduate Student, Department of Radiotherapy, Regional Institute of Medical Sciences, Imphal, Manipur, 795004-India	Professor and Head of the Department, Department of Radiotherapy, Regional Institute of Medical Sciences, Imphal, Manipur, 795004-India	Professor, Department of Pathology, Regional Institute of Medical Sciences, Imphal , Manipur, 795004-India

ABSTRACT *Mucoepidermoid carcinoma (MEC) is a tumour of major and minor salivary gland, arising most commonly from parotid gland followed by minor salivary glands. Rare case reports of mucoepidermoid carcinoma arising from maxilla, maxillary sinus, eustachian tube, bronchi, breast and thyroid were reported. We report a rare case of mucoepidermoid carcinoma of maxillary sinus in a patient with Hepatitis B positivity. The purpose of this review is to discuss that in any maxillary carcinomatous swelling, mucoepidermoid carcinoma could be a differential diagnosis. The location of the tumour close to vital structures makes the surgeon and radiation oncologist difficult to decide on the treatment planning.*

INTRODUCTION

Mucoepidermoid carcinoma is the most common malignant neoplasm of salivary glands occurring more commonly in the parotid gland followed by minor salivary glands. The palate was the most common site for minor salivary gland involvement, accounting for 41.1% of intraoral lesions.[1,2] Atypical sites of mucoepidermoid carcinoma includes subglottis, lung, skin, eustachian tube, breast, thyroid and thymus.[3-9] The most frequent histologic type in paranasal sinus carcinoma were squamous cell carcinoma, followed by adenoid cystic carcinoma, undifferentiated carcinoma, adenocarcinoma and mucoepidermoid carcinomas.[10] At present there is no uniformly accepted grading system for mucoepidermoid carcinoma.[11] Histologically, mucoepidermoid carcinoma is classified into three types of malignancy-low, intermediate, and high grade using five histopathological features namely intracystic component, neural invasion, necrosis, mitosis and anaplasia.MEC is a highly aggressive tumor, its low-grade counterpart glandsusually demonstrates a more benign nature.[12,13]

CASE REPORT

A 33year old male presented to the Department of Radiotherapywith the complaints of swelling on the inner aspect of left side of the face since 2 months. The swelling was gradual in onset and was progressive in nature. He gives past history of visiting a dentist for tooth extraction 1 month back. The swelling started increasing in size following extraction and was associated with foul smelling discharge and halitosis. Patient had history of smoking, alcohol and betel nut chewing since 20 years. On general examination the patient was well built and well nourished. His vital signs were in normal limits. On external examination of face (Fig.1) there was facial asymmetry of the left side.

Oral cavity examination revealed (Fig. 2) carries of the all tooth and a single irregular swelling was seen at on the palatal alveolus in relation to left inferior wall of maxilla. Over the hard palate, a minimal swelling was noticed extending till midline, the associated palatal mucosa was erythematous. On palpation the swelling was non-tender, firm, not fluctuant, not mobile, compressible, and not reducible. An ulcer was also noted on the buccal alveolar swelling. The ulcer was irregular, 5 mm in its greatest extension, the floor was erythematous, margins were sloppy, mildly tender and its base was indurated. Cervical lymphadenopathy was absent clinically. The patient`s Hepatitis B status was found to be positive. Ultrasonography of abdomen showed hepatosplenomegaly with coarse liver echotexture and enlarged portal vein suggesting parenchymal liver disease with portal hypertension. CT scan of abdomen revealed hepatic parenchymal disease with portal hypertension and hepatosplenomegaly.

Panoramic Radiograph (Fig.3) revealed bone loss in maxillary tuberosity area which was extending to the maxillary sinus and floating tooth appearance. CECT of face (Fig.4) revealed heterogeneously enhancing ill-defined soft tissue opacity was seen in the posterior half of left maxillary sinus associated with bony erosion of the medial , inferior and lateral wall of posterior-inferior part of left maxillary sinus and antrum. The lesion was seen infiltrating laterally into the ipsilateral infratemporal fossa and buccal space. Post extraction tooth defect was seen on left upper molar. The bony nasal septum was centrally located, nasal turbinates and meati were normal. Bilateral ethmoid, sphenoid and frontal sinuses are clear and well pneumatized. Radiographic findings suggested that the lesion was extensive, aggressive, osteolytic with its epicenter within the maxilla.

Volume : 6 | Issue : 11 | November 2016 | ISSN - 2249-555X | IF : 3.919 | IC Value : 74.50

Histopathological examination of the swelling (Fig.5, 6) revealed a tissue partially lined by stratified squamous epithelium underneath was a tumour composed of intermediate cells having round to oval nucleus with scant basophilic cytoplasm mixed with epidermoid cells having abundant eosinophilic cytoplasm and occasional cyst. Papillary process projecting into the cyst lumina were noted. Occasional tumour giant cells were seen in the hyalinised stroma. Areas of necrosis were also noted. Features are of Mucoepidermoid carcinoma - low grade. The radiograhic evidence of complete destruction of maxillary sinus along with histological features of low grade mucoepidermoid carcinoma made us to reach final diagnosis of low grade mucoepidermoid carcinoma of maxillary sinus.

DISCUSSION

Mucoepidermoid carcinoma are more common in females than in males with a ratio of 3:1 but maxillary sinus non-squamous cell carcinoma and primary central mucoepidermoid carcinoma have a male predilection. Mucoepidermoid carcinoma is the most frequent malignant salivary gland neoplasm in children. [14,15]

MEC of maxilla usually presents as a painless swelling. Pain, paresthesia, numbness and tooth mobility are usuallyoccasional and late findings.[17]

RadiologicallyMEC can present with exhibit a unilocular or multilocular radiographic lesions, mixed radiopaque-radiolucent lesions, destructive osteolytic lesions with illdefined margins, and radiolucency with calcifications or new bone formation and unusual ground glass appearance.[13,18]

The tumour is histologically comprised of a mixture of mucus cells, intermediate cells and squamoid or epidermoid cells. This tumor may also demonstrate clear cell, oncocytic or columnar cells. Mucoepidermoid carcinoma are graded by most using three tiers: low, intermediate and high grade. The three most popular grading systems are: the AFIP grading system, modified Healey system, and the Brandwein system.[19]

Melnick M et al conducted to determine whether hCMV would be important to the pathogenesis of mucoepidermoid carcinoma (MEC) and was proven that there is a causal relationship between CMV and mucoepidermoid carcinoma. There is insufficient data to establish a relationship between Hepatitis B viral infection and mucoepidermoid carcinoma in this patient.[20] Imaging studies, including CT, should be part of the early clinical management to determine if a neoplasm is present.[21]

Diagnostic workup should include MRI imaging, with or without intravenous contrast since MRI imaging is highly sensitive indetermining the borders and infiltration patterns, as well as perineural invasion to the skull base and meningeal layers.

The standard treatment for low-grade MEC with clinic stage (T1 or T2; N0) is surgical excision. For highgrade tumors, surgery and radiotherapy is recommended.The cure rate after treatment for MEC with minor salivary gland involvement 78,6% for low-grade MEC and 50% for high-grade MEC.[22] The reported overall 5 year survival for MEC ranges from 92 to 100% for low grade tumors, 62–92% for intermediate grade tumors, and 0–43% for high grade tumors.[19] High-grade tumours are highly aggressive and regional lymph node spread is common. The low-grade variant usually demonstrates a favourable outcome, but it is important to note that metastasis may also be present.[23]

Ozawa H et al conducted a study in 43 patients with mucoepidermoid carcinoma in head and neck region with showed the 5-year overall and disease-free survival rate was 62.3 and 57.2% , the study also concluded that the patient's age and treatment method is the prognostic parameter.[24]

Mucoepidermoid carcinoma is a radioresistant tumour, however a rare case of mucoepidermoidtumour treated with low, palliative doses of radiotherapy, resulting in partial response and long term control for more than four years.[25]

Figure 1: Photograph of the patient showing left side facial asymmetry

Figure 2: Photograph of the patient`s oral cavity showing growth in the left palatal region

Volume : 6 | Issue : 11 | November 2016 | ISSN - 2249-555X | IF : 3.919 | IC Value : 74.50

Figure 3 :Panoramic radiograph showing floating tooth appearance

Figure 4: CT scan image of face showing heterogeneously enhancing ill-defined soft tissue opacity was seen in the posterior half of left maxillary sinus

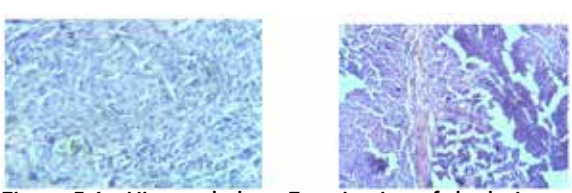

Figure 5,6 : Histopathology Examination of the lesion.

REFERENCES

1. Qureshi SM, Janjua OS, Janjua SM. Mucoepidermoid Carcinoma: A Clinico-Pathological Review of 75 Cases. Int Journal of Oral & Maxillofacial Pathology 2012;3(2):05-09.
2. Bharathi U, Mahesh MS, Lingaraju N, Basappa S, Kalappa TM. Mucoepidermoid carcinoma of palate: A case report. IJSS Case Reports & Reviews 2014;1(7):3-5.
3. Monin DL, Sparano A, Litzky LA, Weinstein GS. Low grade mucoepidermoid carcinoma of the subglottis treated with organ-preservation surgery. *Ear Nose Throat J 2006;85(5):332-6*
4. Heitmiller RF, Mathisen DJ, Ferry JA, Mark EJ, Grillo HC. Mucoepidermoid lung tumours. Ann Thorac Surg 1989;47(3):394-9.
5. Gallager HS, Miller GV, Grampa G. Mucoepidermoid carcinoma of skin. Cancer 1959;12: 286-288.
6. Kawamoto A, Katori Y, Honkura Y, Ogura M, Oshima T, and Kobayashi T. Osseous Hamartoma Arising from the Eustachian Tube. Clin Pract 2013; 3(2):65-67.
7. Pia-Foschini M, Reis-Filho JS, Eusebi V, Lakhani SR. Salivary gland-like tumours of the breast: surgical and molecular pathology. J Clin Pathol 2003;56(7):497–506.
8. Mizukami Y, Matsubara F, Hashimoto T, Haratake J, Terahata S, Noguchi M, Hirose K. Mucoepidermoid carcinoma of thyroid . Cancer 1984;53:1741-5.
9. Noda T, Higashiyama M, Oda K, Higaki N, Takami K, Okami J et al. Mucoepidermoid carcinoma of the thymus treated by multimodality therapy: a case report. Ann Thorac Cardiovasc Surg 2006;12(4):273–278.
10. Dulguerov P, Jacobsen MS, Allal AS, Lehmann W, Calcaterra T. Nasal and paranasal sinus carcinoma: are we making progress? A series of 220 patients and a systematic review. Cancer 2001;92(12):3012–3029.
11. Kolude B, Lawoyin JO, Akang EE. Mucoepidermoid carcinoma of the oral cavity. J Natl Med Assoc. 2001;93(5):178–184.
12. Pires FR, Almeida OP, Araújo VC, Kowalski LP. Prognostic Factors in Head and Neck Mucoepidermoid Carcinoma. Arch Otolaryngol Head Neck Surg. 2004;130(2):174-180.
13. Suresh D, Raviraj J, Vijaykumar B, Suman SV, Suneel KV, and Amrutha K. Central mucoepidermoid carcinoma of the maxilla with unusual ground glass appearance and calcifications: A case report. Imaging Sci Dent 2014 ; 44(2): 161–164.
14. Daryani D, Gopakumar R, and Nagaraja A. High grade mucoepidermoid carcinoma of maxillary sinus. J Oral Maxillofac Pathol 2012; 16(1):137–140.
15. Hicks J, Flaitz C. Mucoepidermoid carcinoma of salivary glands in children and adolescents: assessment of proliferation markers. Oral Oncol 2000;6:454–460.
16. Myers LL, Nussenbaum B, Bradford CR, Teknos TN, Esclamado RM, Wolf GT. Paranasal sinus malignancies: an 18-year single institution experience. Laryngoscope 2002;112:1964–1969.
17. Rathore AS, Ahuja P, Chhina S, Ahuja A. Primary intraosseous mucoepidermoid carcinoma of maxilla. J Oral Maxillofac Pathol 2014; 18(3): 428–431.
18. Sherin S, Sherin N, Thomas V, Kumar N, Sharafuddeen KP. Central mucoepidermoid carcinoma of maxilla with radiographic appearance of-mixed radiopaque–radiolucent lesion: a case report. Dentomaxillofac Radiol. 2011; 40(7): 463–465.
19. Seethala RR. An Update on Grading of Salivary Gland Carcinomas. Head and Neck Pathol 2009;3(1):69–77.
20. Melnick M, Sedghizadeh PP, Allen CM, Jaskoll T. Human cytomegalovirus and mucoepidermoid carcinoma of salivary glands: cell-specific localization of active viral and oncogenic signaling proteins is confirmatory of a causal relationship. Exp Mol Pathol 2012;92(1):118-25.
21. Mardi K, Singh S. Primary mucoepidermoid carcinoma of maxillary sinus- a rare case report. Int J Otorhinolaryngol 2008;10(1):1-4
22. Sepúlveda I, Spencer L, Platin E. Sinonasal Mucoepidermoid Carcinoma: a case report and literature review. Int J Odontostomat. 5(3):304-308, 2011.
23. Goode RK, Auclair PL, Ellis GL. Mucoepidermoid carcinoma of the major salivary glands: clinical and histopathologic analysis of 234 cases with evaluation of grading criteria. Cancer 1998;82:1217–1224.
24. Ozawa H, Tomita T, Sakamoto K, Tagawa T, Fujii R, Kanzaki S, Ogawa K, KameyamaK, Fuji M. Mucoepidermoid Carcinoma of the Head and Neck: Clinical Analysis of 43 Patients. Jpn J Clin Oncol 2008;38(6):414–418.
25. Vulpe H, Giuliani M , Goldstein D, Perez-Ordonez B, Dawson LA, Hope A. Long term control of a maxillary sinus mucoepidermoid carcinoma with low dose radiation therapy: a case report. Radiat Oncol 2013; 8:251.