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Case Report

KERATINIZING SQUAMOUS CELL CARCINOMA- A RARE CASE REPORT

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ABSTRACT:

Keratocystic odontogenic tumor (KCOT) is still a dilemma in field of medical science because of its biological behavior and transformation of this benign odontogenic tumor to a carcinomatous type, which is called as Odontogenic carcinoma. We are presenting here a case of squamous cell carcinoma occurring in recurrent keratocystic odontogenic tumor of temporal region. The rarity of this article is the location of lesion which is rarest at temporal region. Malignant transformation of these KCOT's has been reported only in 15 cases in literature, majority occurring in jaws but no case reported for lesion of temporal region. The pathogenesis of the tumor, the biologic progression, overall clinical and histopathological features of this rare malignancy and its management is reported and discussed.

KEY WORDS: Keratinizing squamous cell carcinoma, Odontogenic carcinoma, temporal region

INTRODUCTION:

Keratocystic Odontogenic tumor is still a unique pathological entity due to its anatomical & biological behavior since it description by Phillipsen in 1956.1 Squamous cell carcinoma (SCCA) arising from the wall of KCOT is a rare which occurs mainly in jaw bones called as keratinizing squamous cell carcinoma (odontogenic carcinoma) as first described by Loos in 1913.2,3 Management of these lesion at rare anatomic location possess a challenge for the surgeon.

CASE REPORTS:

A 66 year old female patient reported to our Department of Oral & Maxillofacial Surgery complaining with a slow growing painless swelling in Right side temporal region from past 1 month. According to her previous reports she had a history of Enucleation of KCOT in Right side posterior region with extraction of 46, 47 and 48, 5 years back. There was a recurrence of Multilocular KCOT again 2 years back for which she underwent Hemimandibulectomy with disarticulation of the condyle on right side till 32. She had no history of tobacco or illicit substance use. Maxilla was edentulous and atrophic. On examination there was a painless swelling measuring about 5x4 cm , round in shape with well defined borders in right temporal region which was fixed to the temporalis muscle . The swelling was non tender, non fluctuant and non pulsatile. Her medical history was non contributory. No palpable local or regional lymphadenopathy was seen.

Radiological examination included CT brain. There was a hypodense swelling involving soft tissue on the right side temporal region. No involvement of the bone was seen. On Aspiration biopsy, we got a

dark yellow straw coloured fluid. After a complete clinical, radiological and biopsy examination, provisional diagnosis of Recurrent KCOT was made. Wide local surgical excision of the tumor in general anaesthesia was performed . The completely resected specimen was sent for histopathological examination, which was suggestive of well differentiated keratinizing SCCA of the temporal region. In some areas the lining showed increasing dysplastic change, transforming ultimately into invasive keratinizing SCCA. Postoperative ultrasonography of abdomen was done which was negative to rule out any primary site of tumor. There was transient facial nerve palsy which has recovered in 3 months, otherwise postoperative recovery was uneventful. There was presence of temporal hollowing but considering the age of the patient it was of minimal facial esthetic concern. Patient was sent for prophylactic radiotherapy. In follow-up of 6 consecutive months, patient is doing well and there was no recurrence of lesion or lymphydenopathy seen till present.

DISCUSSION:

Van der Wal et al 4 reported that the presence of keratinization in the cystic lining is a risk factor for malignant changes and Browne et al. indicated that odontogenic cysts with keratinization are often prone for malignant changes than non-keratinizing cysts. Eversole et al 5 in 1975 reported out of 32 cases of central carcinoma, 75% were often associated with a cyst. In 1975, Gardner 6 proposed certain criteria for diagnosis of SCCA arising in an odontogenic cyst. They are microscopic transition area from benign cystic epithelial lining to invasive SCCA, there must be no carcinomatous changes in the overlying epithelium and no source of carcinoma in the adjacent structures. A 4th criterion added by Waldron[7] was the possibility that the lesion represents a metastasis from a distant tumor must be ruled out by physical and radiological examination and the subsequent clinical course. Several features of this case support Waldron's criteria. There was no evidence of any primary tumor elsewhere in the body. There were no signs of paresthesia probably because the malignant changes were confined within the cystic lining. Discovery of SCCA arising in this KCOT was unexpected.

MRI is an investigation of choice for soft tissue swellings but since our patient was having financial crisis it could not be done. Treatment of choice for these lesions is surgery with wide margins with 1 to 1.5 cm clearance of unaffected tissue. Intraoperative Frozen section analysis can be done to achieve tumor free surgical margin if the diagnosis of malignancy is made before the treatment procedure. Addressing of the nodes has to be done if there is local or regional lymph node metastasis with appropriate type of neck dissection. In this case, since the patient did not have any local or cervical lymphadenopathy hence neck dissection was not done. Radiation therapy can be given as an adjuvant as in this case. Bone scintigraphy can be done to rule out any metastatic bone diseases. In general, the prognosis is poor and metastasis to cervical lymph nodes is observed in up to 50% of cases in a well established SCCA. Two year survival rate of patients has been reported in 53%8.

CONCLUSION:

This article analyzes the origin, clinical and histopathological features, and surgical outcome of a malignant transformation of a KCOT of the temporal region in to SCCA. It illustrates the importance of adequate microscopic investigation of all recurrent excised cysts and their appropriate management to improve the prognosis of a patient.

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