



ADENOID CYSTIC CARCINOMA OF THE BASE OF TONGUE PRESENTING WITH LIVER METASTASIS AFTER 7 YEARS

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ABSTRACT

Adenoid cystic carcinoma (ACC) is an unusual malignancy from the base of the tongue. Metastasis from minor salivary gland malignancy including ACC usually is seen at the time of diagnosis or within one year of completion of treatment as recurrences. We present here a case of ACC of the base of tongue, who following successful surgical resection and radiotherapy presented with liver metastasis after 7 years. ACC is not so common malignant tumour of the base of tongue and distant metastasis from it can manifest very late.

INTRODUCTION

Adenoid cystic carcinoma (ACC) is a neoplasm of the salivary gland and it accounts for approximately 5-10% of all salivary gland neoplasms [1]. The most frequent localization is in the parotid gland (30%), followed by the sub mandibular gland, 40% are found in minor salivary glands and 1% in the sub lingual gland [2]. The frequency of occurrence of ACC in the tongue is 19.8% with almost 85% of all tongue cases are reported from the base of tongue [3].

The ACC are generally slow growing tumours and spread to nearby structures by peri-neural route. Hematogenous spread is more common than lymphatic spread [4]. Distant metastasis is usually seen in the lungs but may also occur in brain, bone, liver, thyroid and spleen [5].

We present here a case of ACC of the base of tongue, who following successful surgical resection and radiotherapy presented with liver metastasis after 7 years.

CASE REPORT

A 29 years old female presented with the complaints of swelling on the left side of the neck for 3 years and difficulty in swallowing for 1 year. On examination, the swelling measured 10.5cm X 6 cm on the left side of the neck, and it was non-tender and hard in consistency. Indirect laryngoscopy revealed a growth on the posterior 3rd of the tongue. Endoscopy showed ulceroproliferative growth with induration in tongue, base of tongue and vallecula. Computed tomogram (CT) scan of the oral cavity and neck showed lobulated lesion in left side of base of tongue intruding inferiorly to the root of vallecula and underlying muscles. Punch biopsy from the growth in base tongue showed features of adenoid cystic carcinoma. The clinical staging was T4N3M0. The patient underwent total glossectomy with neck dissection. Histopathological examination showed adenoid cystic carcinoma (Figure 1) involving muscle tissue. Further the patient received external beam radiotherapy (EBRT) with 56 Gy to bilateral face and neck with 60 CO X-rays. The

patient was on regular follow up. Seven years after primary surgery followed by EBRT, the patient presented with an abdominal swelling. The patient was icteric and abdominal examination revealed solid mass in the epigastric and umbilical region extending to right lumbar region. Contrast CT abdomen revealed fairly large hypodense, multilobulated, conglomerate hepatic mass almost entirely involving the right lobe sparing only a part of the peripheral aspect. Liver function tests revealed increased

bilirubin and liver enzymes and decreased albumin. Tumour markers of both CA 125 and Carcinoembryonic Antigen (CEA) were elevated but alphafeto protein (AFP) was found to be within normal range. Fine Needle Aspiration Cytology (FNAC) from liver favoured metastatic adenoid cystic carcinoma (Figure 2).

Since the liver lesion was large, so, surgical option was not considered and the patient was referred for palliative care only.

Figure 1. Photomicrograph showing nests and columns of cells arranged around glands like spaces. (H&E,400X)

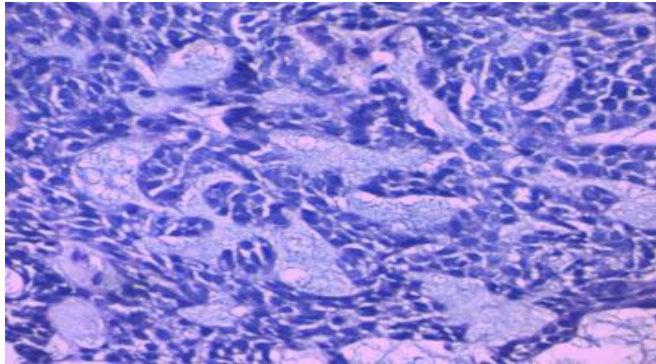
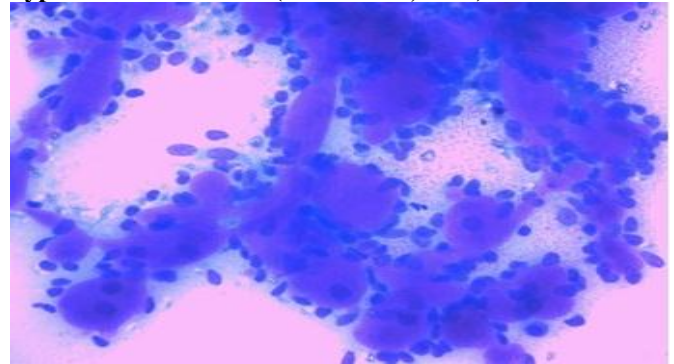


Figure 2. Finger like fragments of hyaline stroma between the tumour cells with round to oval hyperchromatic nuclei (MGG stain,400X)



DISCUSSION

Adenoid cystic carcinoma (ACC) is a malignant neoplasm that accounts for 1-2% of all head and neck cancers [1]. Local recurrence of ACC is common despite aggressive surgical resection due to higher chances of tissue infiltration and nerve spread [6]. However, late metastasis is rare. ACC are generally neurotropic, that is why its spreads through nerves are common and thus during resection, frozen section is suggested [7]. ACC has a very slow growth and tends to have a protracted course [8,9]. ACC also shows late distant metastasis in rare events [10], like in our case. The sites of metastasis are usually the organ containing the first capillary bed [11]. So, patients with ACC should be advised long periods of follow-up. The stage and tumour location are factors in ACC which are prognostically significant [12]. Hepatic metastasis of ACC occurs when there is dissemination of the disease to multiple organ sites, and only hepatic metastasis is very rare [13]. In our patient tumour markers CA 125 and CEA were elevated. CEA tends to rise in patients with colon and rectal cancer but it may non-specifically increase in any patients with abdominal mass causing peritoneal irritation. Same is with CA-125, which is specific for ovarian cancer but may also increase in any abdominal mass and AFP does not tends to increase in hepatic metastasis. All these are reflected in tumour marker findings of this patient. Metastasis in lymph node at presentation is rare but when present have bad prognosis [14]. Generally surgical resection followed by radiotherapy is used for better result outcome and use of radiation improves local control and disease free survival. This

patient received radiation and did not have any local recurrence after long time of follow-up (7 years). However, this patient developed metastasis in the liver after a long time following treatment and as the first and only organ. Also, she did not showed any evidence of distant metastasis to other organs or local recurrence. Surgical options were not explored as large area of liver was involved. It is said that distant metastasis at the cellular level could have occurred many years prior to clinical presentation.

CONCLUSION

ACC is not so common malignant tumour of the base of tongue and distant metastasis can manifest very late. A long term follow up is necessary after a successful completion of treatment even with disease free or recurrence free condition at the 5-years follow-up period.

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CONFLICT OF INTEREST

The authors declare that they have no conflict of interest.

STATEMENT OF HUMAN AND ANIMAL RIGHTS

All procedures performed in human participants were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. This article does not contain any studies with animals performed by any of the authors.

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