

# Metastasis of Adenoid Cystic Carcinoma of Buccal Mucosa to Lungs - A Case Report with Review of Literature

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## ABSTRACT

**Introduction:** Adenoid cystic carcinoma (ACC) is a malignant neoplasm that accounts for 1-2% of head and neck malignancies and 10-15% of all salivary gland malignancies. Peak incidence is in 6<sup>th</sup>-7<sup>th</sup> decades of life with a slight female preponderance. ACC are deceptive malignancies that show slow growth and local invasion with recurrences many years after diagnosis. Upto 50% of these tumors occur in the intraoral minor salivary glands usually hard palate. Buccal mucosal tumors are relatively rare. Incidence of distant metastasis in salivary gland cancer is relatively low and has poor prognosis.

**Case Report:** A 50 year male patient, diagnosed case of ACC buccal mucosa presented with chief complaints of cough, breathlessness and fever. Physical examination and investigations like X- ray chest and CT Thorax, raised suspicion of lung metastasis which was confirmed by FNAC Lung revealing metastatic carcinomatous deposits of Adenoid Cystic Carcinoma.

**Conclusion:** ACC is a malignant tumor of salivary gland with a deceptively benign histologic appearance characterized by indolent, locally invasive growth with high propensity for local recurrence. Distant metastasis mostly appears to be delayed after primary diagnosis and therapy. Typical symptoms of metastasis are not very well known and are non-specific. Radiological screening of every patient with suspicion of salivary gland tumour metastasis should be done. Detection of distant metastasis at initial evaluation may alter the selection of therapy and is therefore very important. Patients with Adenoid Cystic carcinoma should have an X-ray or CT scan of the chest to exclude the possibility of distant metastasis.

**Keywords:** Adenoid Cystic Carcinoma, Metastasis, Lungs

## INTRODUCTION

Adenoid cystic carcinoma (ACC) is a malignant neoplasm of the salivary glands. It was first described as cylindroma by Billroth in 1856.<sup>1</sup> ACC constitute less than 1% of all head and neck malignancies with 50% of all ACCs occurring intraorally, commonly in the hard palate. Other less common intraoral sites include the lower lip, retro molar/tonsillar pillar region, sublingual gland, buccal mucosa and floor of the mouth.<sup>2</sup> The peak incidence is in sixth and seventh decades of life with a slight female preponderance.<sup>3</sup>

ACC is an indolent tumor but grows relentlessly. It is typically unencapsulated. This infiltrative capacity is the hallmark of this carcinoma because of which multiple local recurrences are common. ACC progresses slowly with wide perineural invasion into the adjacent nerves, suggesting the presence of strong neurotropism for the tumor.<sup>4</sup>

ACC occasionally invades regional lymph nodes by direct extension, but true embolic lymph node metastasis is rare. However, distant metastasis to organs other than lymph nodes is common, particularly late in the course of the disease, the lungs being the primary site followed by bones, the liver and

the brain.<sup>5-7</sup> We present a case of bilateral lung metastasis in a male patient with buccal mucosa swelling diagnosed as ACC and review the literature.

## CASE REPORT

A 50 year male patient, an already diagnosed case of Adenoid cystic carcinoma buccal mucosa presented to Guru Gobind Singh medical college, Faridkot with chief complaint of cough for 4 months, breathlessness for 2 months and fever since 20 days.

Patient was diagnosed ACC in 2005 at PGI, Chandigarh where he presented with complaint of swelling on the buccal mucosa (left side) for the last 9 years which started spontaneously and then showed a rapid increase in size over one month. A diagnosis of ACC was made on biopsy. Selective neck dissection and flap reconstruction were performed, followed by 6 cycles of concomitant radio-chemotherapy.

Now the Cough was productive with mucoid expectoration, at times blood tinged for the last 4 months. Physical examination showed good general condition, poor oral hygiene. Pallor and clubbing was present. There was history of significant weight loss with loss of appetite.

Chest X ray P-A view showed multiple cannon ball opacities (Figure-1).

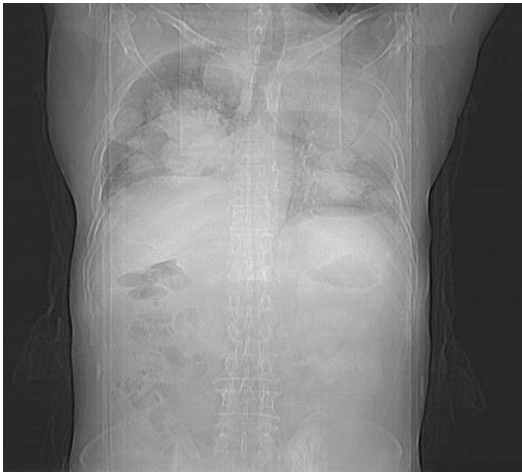
A CECT scan of Thorax and upper abdomen revealed multiple, inhomogeneously enhancing, pleural based and parenchymal soft tissue density lesions of variable sizes in both lung fields with few of them showing areas of necrosis within suggesting metastatic deposits. The largest one in right lower lobe measures 11.3x8.8cms in axial plane and largest one in left upper lobe measures 10.7x7.8cms in axial plane. A few small mediastinal lymph nodes and multiple osseous metastatic deposits were seen (Figure-2).

Fine needle aspiration cytology of the lung was performed. Smears showed collection of atypical cells exhibiting high N:C ratio, round hyperchromatic nuclei along with the presence of hyaline globules. In the background inflammatory cells consisting of neutrophils, lymphocytes and macrophages were seen. These features suggested the Cytological diagnosis of Metastatic carcinomatous deposits Of Adenoid cystic carcinoma

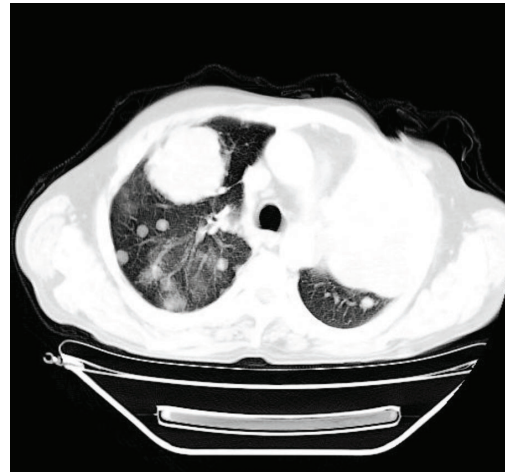
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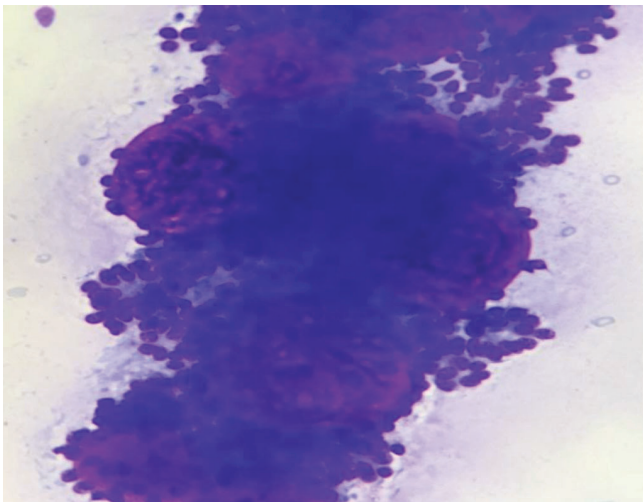
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**Figure-1:** X-ray chest PA view shows multiple cannon ball opacities.



**Figure-2:** CT Thorax showing multiple metastatic deposits in both the lung fields



**Figure-3:** Photomicrograph (x10) showing basaloid tumor cells adherent to hyaline globules

(Figure- 3).

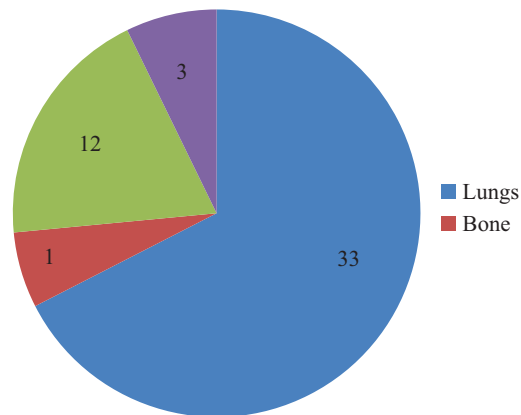
Patient was referred to Department of Oncology for further management.

## DISCUSSION

Adenoid Cystic Carcinoma (ACC) is a clinically and pathologically well-defined entity that has been described in the literature extensively. WHO definition of ACC is, "A basaloid tumor consisting of epithelial and myoepithelial cells in various morphological configurations including tubular, cribriform and solid patterns. It has a relentless clinical course and, usually, a fatal outcome."

As for the primary location of the ACC in a total of 193 subjects in a study, the most common area was palate, accounting for 57% of cases, corresponding to 110 patients; followed by tongue with 31 cases (16%); in oral mucosa with 28 cases (14.5%); in fourth position was floor of the mouth with 16 cases (8.3%); and lastly mandible and lip.<sup>8</sup> Toida et al affirm in their study of 10 patients that the most common location is in palate, representing 70% of the cases, followed by oral mucosa, a finding that coincides with the study conducted by Dhanutai et al.<sup>9,10</sup>

ACC arises from the mucous-secreting glands. These mucous-secreting tumors are confined to structures derived from the foregut (that is, the parotid, submandibular and sublingual



**Figure-4:** Location of distant metastasis

glands, and the mucous glands throughout the upper respiratory tract).

Classically, ACC has a distinct natural history characterized by slow and indolent growth, with late development of metastasis and often distant recurrences. Lymphatic spread is rare. Distant metastases can occur to the lungs and bones. The susceptibility for developing distant metastasis from a salivary gland tumor depends on four factors: time from initial diagnosis, histotype, tumor stage and location of the primary tumor. In a review of the literature 8.1% of 835 patients with minor salivary gland cancer developed distant metastasis.

Metastasis was evaluated in all of the 193 patients in a study, finding a total of 96 metastases, 48.96% of which were at the local level, whereas 51.04% were distant metastases. With respect to the topographic location of the 49 distant metastases, 33 were located in the lungs, 12 in the lungs and bone, 3 in the brain and only one in the bone (Figure 4).<sup>8</sup>

## CONCLUSION

Distant metastasis in head and neck cancer, especially in salivary gland malignancies, is a rare course of disease. Distant disease mostly appears to be delayed after primary diagnosis and therapy. Consequently, typical symptoms of metastasis are not very well known and are often nonspecific, especially in elderly patients with other co-morbidities. The detection of distant metastasis at initial evaluation may alter the selection of therapy and is therefore very important. As the appearances

of loco-regional tumor recurrence or distant metastasis are prognostic factors for tumor progression, radiological screening of the lung has to be considered in patients suspected of having salivary gland tumor metastasis.

Each patient should have a CT scan of the chest. An abdominal ultrasound should be performed. Special investigations, e.g. bone scintigraphy or abdominal CT scan, are indicated in cases with clinical symptoms that raise suspicion of metastasis.

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