

CASE REPORT

Cervical Lymph Node Metastasis: Unusual Presentation of Adenoid Cystic Carcinoma - Diagnosed By FNAC

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Abstract:

Adenoid cystic carcinoma (ACC) is a rare neoplasm that usually arises from minor salivary glands. It is characteristically locally infiltrative, exhibiting perineural invasion, has a tendency for local recurrence and prolonged clinical course. A 60 year old male, chronic smoker presented with swelling of the left cervical lymph node since two months. Examination revealed a solitary firm, non tender, non mobile left cervical swelling measuring 2 x 1 cm. Fine Needle Aspiration Cytology (FNAC) was done from the cervical lymph node. The diagnosis of metastatic deposits of ACC was given. Detail examination of the oral cavity revealed a small swelling at the floor of the mouth. Biopsy of the swelling confirmed ACC on histopathological examination. An unusual feature of adenoid cystic carcinoma is the low incidence of metastases to regional lymph nodes. The case is presented to highlight its unusual presentation and utility of FNAC in rapid diagnosis.

Keywords: Adenoid Cystic Carcinoma, FNAC, Salivary Gland

Introduction:

Adenoid cystic carcinoma (ACC) is a rare neoplasm that usually arises from salivary glands. It accounts for less than 1% of all head and neck malignancies and about 10% of all salivary gland tumours. However, it is the commonest malignant tumour of the minor salivary gland [1,2]. It is also seen in other rare locations which include the aero-digestive tract, lacrimal glands and adnexal skin glands. Most ACC patients are in their fifth and sixth decades of life, with a slight female

preponderance. It is prone to local infiltrative growth, often exhibiting perineural invasion, with a tendency for local recurrence and prolonged clinical course. Lymph node metastasis is less often noted. Cervical metastasis is rare and occurs in less than 5% of patients. Hence many institutions consider this tumor as 'low risk' for lymph node metastasis and do not perform neck dissection if clinicoradiographically node negative. The importance of presenting this case is for the prior detection of cervical metastasis of ACC by FNAC, which is rare. This cytodiagnosis helped in early detection of the lesion and planning the appropriate surgical management to improve the prognosis.

Case Report:

A 60 year old male patient, a chronic smoker, presented with enlarged left cervical lymph node in anterior triangle of the neck since 2 months. He also complained of mouth ulcer on the left side of the floor of the mouth since 4-5 months. There was no history of fever, weight loss or dysphasia. Family and past history were not significant. Examination revealed a solitary left cervical swelling in anterior triangle of neck measuring 2x1 cm which was firm, fixed and non tender. FNAC was done from the cervical lymph node. Smears were dried, fixed and stained with leishman's and H&E stains. Smears were cellular, composed of multiple hyaline spherical globules of varying size surrounded by tumour cells. The

tumour cells were round having uniform round to oval hyperchromatic nuclei and scanty cytoplasm (Fig. 1). The background showed few lymphocytes. The H&E stained slide was paucicellular. Re-examination of the oral cavity showed a small swelling beneath the ulcer on the floor of the mouth. Biopsy was done from this site. Tumour

cells were arranged in cribriform pattern, nests and cords. The individual cells were small, round arranged around gland like spaces filled with eosinophilic material (Fig. 2). This confirmed the diagnosis of ACC of the minor salivary gland of the floor of the mouth.

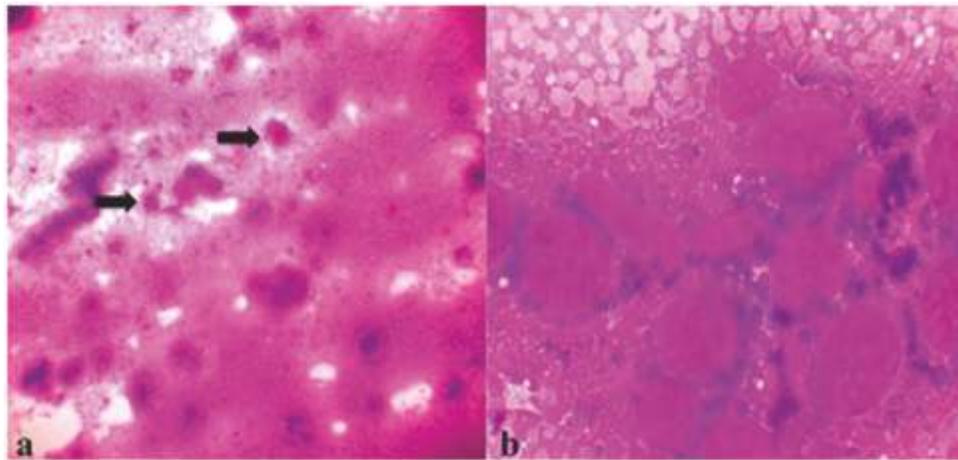


Fig. 1a: FNAC Smear Showing: Globules of Hyaline Material (Arrow) (Leishman, 100X)

Fig. 1b: FNAC Smear Showing: Hyaline Material Surrounded by Tumour Cells (Leishman, 400X).

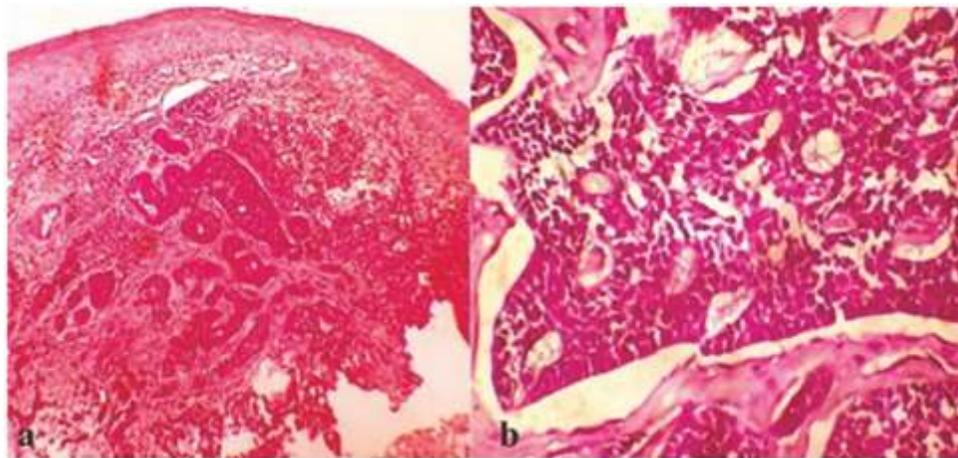


Fig. 2a: Photomicrograph of Biopsy from Primary Site showing: Tumour Cells arranged in Cribriform Pattern, Nests and Cords (H & E 100X)

Fig. 2b: Photomicrograph of Biopsy from Primary Site showing: Small Round Tumour Cells around Gland like Spaces Filled with Eosinophilic Material (H&E, 400X).

Discussion:

ACC was first described by Bilroth in 1859, using the term “cylindroma”. At present ACC accounts for 29.6% of tumours of minor salivary gland with bimodal age distribution (mean 40 years).

Grossly, it usually has a solid appearance and an infiltrative pattern of growth. Cytological diagnosis of adenoid cystic carcinoma is based on findings of hyaline spherical globules of varying size surrounded by tumour cells. The individual cells are small and have round or ovoid nuclei and a narrow rim of cytoplasm. The nuclei show little variation in the size and shape with readily visible nucleoli. The homogenous acellular material is referred to as the main cytological feature, which was the key diagnostic feature in the present case. Hyaline globules are seen in variety of salivary gland neoplasms like ACC, pleomorphic adenoma and basal cell adenoma [4]. The hyaline globules are intensely metachromatic on Diff Quik stain and clear to cyanophilic on Papanicolou stain. A true differential diagnostic problem lies in the fact that a pleomorphic adenoma may occasionally present features mimicking classic ACC. In pleomorphic adenoma tumour cells usually form flat sheets rather than multilayered clusters, characteristic of ACC [5]. The cytological identification of ACC rests on adequate sampling and careful inspection of all material to rule out the possibility of benign pleomorphic adenoma and basal cell adenoma. Polymorphous Low Grade Adenocarcinoma (PLGA) of minor salivary gland can also have papillary cribriform or solid growth pattern. This tumour is the most difficult to differentiate from ACC on cytology. The tumour cells of PLGA are small with ovoid nucleus and thin rim of cytoplasm [6]. These cytological features are sometimes distinct enough to allow diagnosis of PLGA.

A homogenous mesenchymal substance similar to that formed by ACC can also be seen in epithelial

myoepithelial carcinoma. The presence of clear cells in aspirate from epithelial and myoepithelial carcinoma permits recognition of most cases. Immunocytochemistry for myoepithelial markers is conclusive.

An unusual feature of adenoid cystic carcinoma is the low incidence of metastasis to regional lymph nodes. Lymphatic spread being very rare, lymphadenopathy is seldom encountered. Lymph nodes, in very extensive cases, may be involved by direct extension [7]. In this case, primary tumour was away from the cervical lymph node, ruling out the possibility of direct spread. Distant metastasis may occur in up to 50% of ACC patients during the course of the disease, with the lungs and liver as the most common sites [8]. Bone metastasis indicates a fulminant clinical course [1]. Advanced tumour stage, solid histological type, presence of nodal metastasis, positive margins and perineural spread points towards a poor outcome.

The behaviour of adenoid cystic carcinoma of the salivary glands has been shown to be unpredictable in terms of local recurrence, distant spread and mortality. Clinically two distinct patterns are noted. Few cases have a relentless fulminating course with early metastasis and fatality within short period of 2-3 years. Most of the other cases have an insidious natural history and long survival period despite of local recurrence. Treatment of adenoid cystic carcinoma includes a complete excision of the local disease followed by post-operative radiotherapy. Detection of cervical lymph node metastasis by FNAC, as in the present case needs neck dissection.

The case is presented to highlight an unusual presentation of ACC of minor salivary gland as cervical lymphnode metastasis, its characteristic cytomorphological appearance and utility of FNAC in rapid diagnosis and planning the surgical management.

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