Case Report

Unusual Presentation of Adenoid Cystic Carcinoma

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Abstract

Adenoid cystic carcinoma is a malignant neoplasm most commonly originating in salivary glands of head and neck region. Its occurrence elsewhere is rare and extension to thyroid even rarer. We report a 27 year old male who was diagnosed to have features of adenoid cystic carcinoma in fine needle aspirate of the thyroid swelling and subsequently confirmed by histopathology. Extensive search failed to reveal a primary. Awareness of cytology of adenoid cystic carcinoma and unusual presentation can help in making a diagnosis on cytology.

Key Words: Adenoid cystic carcinoma, thyroid, metastasis.

Introduction

Adenoid cystic carcinoma accounts for approximately 10% of all salivary gland tumours. It is the most common malignant tumour of submandibular and minor salivary glands. The tumour extends well beyond the visible and palpable limits of the salivary gland region; this infiltrative capacity is the hallmark of this carcinoma. Pain and spreading along nerve sheath is often noted. While adenoid cystic carcinoma is not an uncommon tumour in the salivary glands, its occurrence elsewhere is rare. Extension into the thyroid gland is extremely rare. However, if the cytologic features are in favour of adenoid cystic carcinoma, this possibility should be considered in thyroid aspirates.

Case Report

A 27 year old male came to ENT out patient department with a history of pain in throat, difficulty in swallowing and difficulty in breathing for the past 4 months. On examination, his thyroid was irregularly mildly enlarged. Ultrasonography was advised which revealed an enlarged left lobe showing multiple mixed echoic lesions in the thyroid parenchyma along with calcified focal lesion of 16x14 mm in upper part of the left lobe. Computed tomography (CT) scan study of neck revealed a low-density mass arising from the left lobe of thyroid extending medially into the isthmus and inferiorly extending into the superior mediastinum. The lesion was displacing and compressing the trachea towards the right side and the major vessels of the neck laterally. The trachea did not show evidence of any lesion, thereby ruling out primary from trachea. No vascular encasement was seen. Esophagus was also compressed. A radiological suspicion of thyroiditis / lymphoma was entertained and tissue diagnosis was advised. Fine needle aspiration (FNA) of the thyroid region was done using a 23-gauge needle by non-aspiration technique. Wet fixed and air dried smears were made and stained with Hematoxylin and Eosin (H&E) and May-Grünwald Giemsa (MGG) stains.

Microscopic findings: The smears were cellular and showed mostly syncytial tissue fragments with crowding and overlapping of the cells. There were three-dimensional clusters containing homogenous acellular hyaline material (Fig. 1). The cells were small, round to oval, monomorphic with a high N/C ratio. The nuclei were round with coarsely granular chromatin and inconspicuous nucleoli. The cytoplasm was scant and indiscernible. The acellular hyaline material was PAS positive. A diagnosis of adenoid cystic carcinoma was made. On exploration the mass appeared to be inoperable. A biopsy was taken which confirmed the diagnosis of adenoid cystic carcinoma (Fig. 2). No thyroid tissue was seen in the specimen submitted for histopathology.

The patient was referred for radiotherapy.

Discussion

Adenoid cystic carcinoma is the most common primary in the major and minor salivary glands but can also arise from submucosal seromucinous glands of larynx and trachea and from lung. When it presents outside of these locations, the diagnosis becomes more challenging. It is a slow growing tumour, with a high incidence of distant metastases.
The morphologic features are same, regardless of the site. There are three recognized histologic subtypes: tubular, cribriform and solid. The most common form is the cribriform type. It spreads by direct extension, perineural invasion, and hematogenous route. Lymphatic spread is uncommon.5

Adenoid cystic carcinoma presenting as a thyroid nodule is very rare. Direct extension into thyroid by malignant neoplasm from laryngotracheal complex, although very uncommon, is well documented.2 Although metastatic lesions are not common in the thyroid, renal cell carcinoma, breast carcinoma, endocervical carcinoma and lung carcinoma have been encountered.6 Metastasis from parotid has also been reported.7

In the present case there was a mass in the left lobe of thyroid. On CT scan examination the lesion was extending into the isthmus and into the superior mediastinum inferiorly. It was displacing and compressing the trachea but as such no lesion was present in the trachea. No lesion was detected in the salivary gland. FNA revealed features of adenoid cystic carcinoma. The patient was posted for surgery. Since the lesion was extensive and inoperable when opened by the surgeon only a small biopsy was taken and the lesion was closed. Possibility of metastatic adenoid cystic carcinoma of thyroid from an unknown primary was reported.

The present case highlights the need to be aware of unusual presentation of adenoid cystic carcinoma that may arise in the region of thyroid. Knowledge of these lesions that can present as thyroid mass will help in making a correct diagnosis on cytology.

References
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