

Unusual presentation of metastatic adenoid cystic carcinoma: a challenge in aspiration cytology of the thyroid

B.J. ROCCA^{1,2}, A. BARONE¹, A. GINORI¹, M.R. AMBROSIO¹, A. DISANTO³

¹ Department of Medical Biotechnologies, Section of Pathology, University of Siena, Italy; ² Section of Pathology, Ospedale di Circolo di Busto Arsizio, Presidio Ospedaliero di Saronno, Italy ³ Azienda Ospedaliera Universitaria Senese, Section of Pathology, Siena, Italy

Key words

Fine needle aspiration cytology • Adenoid cystic carcinoma • Metastasis to thyroid • Minor salivary glands

Summary

Introduction. Adenoid cystic carcinoma is a malignant neoplasm most commonly originating in the salivary glands. Its occurrence elsewhere is rare and its metastasis to the thyroid gland has been described only once.

Case report. We describe the case of a 66-year-old man who presented for a swelling in the midline neck of six months duration. A solitary palpable nodule was identified in the isthmic region of the thyroid. Fine needle aspiration of the nodule revealed high cellularity, a partial microfollicle-like pattern and the presence of small hyaline globules. The neoplastic population was composed

of monomorphic cells with basaloid appearance. Thyroid primitivity was excluded on the basis of the negativity for TTF1 and thyroglobulin. As the patient referred an ulcerative lesion of the inferior lip, fine needle aspiration cytology of the lesion was performed, yielding a diagnosis of adenoid cystic carcinoma.

Conclusion. The present case highlights the need to be aware of possible metastatic thyroid localisation of adenoid cystic carcinoma also originating in minor salivary glands of the oral cavity. This is a very rare event, but it should be taken into consideration and clinical and cytological findings must be carefully examined.

Introduction

Adenoid cystic carcinoma (ACC) accounts for 3-5% of all salivary gland tumours and occurs in both minor and major salivary glands¹, the former representing the most common malignancy². It can also arise from submucosal seromucinous glands of the larynx and trachea as well as the lung³. When it presents outside these locations, diagnosis is more challenging. Cytologically, the tumour is characterised by clusters of cohesive monomorphic small round to oval epithelial cells, with a high nuclear/cytoplasmic ratio, round nuclei, coarsely granular chromatin, prominent nucleoli and scant cytoplasm. The clusters surround globules of homogenous, acellular hyaline magenta-stained material. Dispersed naked nuclei are common as well as spherical aggregates, rosette-like groups, papillary structures and solid nests of cancer cells⁴. The tumour often has a relentless clinical course with local recurrences and high incidence of distant me-

tastases, owing to its infiltrative growth⁵. The lungs and central nervous system are sites of predilection of distant metastases⁶⁻⁸. Thyroid metastases from non-salivary gland ACC has been described in rare cases^{6,9,10}. In our extensive review of the literature, we found only one case of salivary gland ACC metastatic to thyroid in a patient with ACC of parotid gland^{6,11}. Herein, we describe the first case of ACC of a minor salivary gland metastatic to the thyroid.

Case report

A 66-year-old man presented for the onset of a swelling in the midline neck lasting six months. The swelling was slowly progressive and was not associated with pain or compressive symptoms. The patient was clinically euthyroid. On physical examination, a solitary palpable nodule was identified in the isthmic region of the

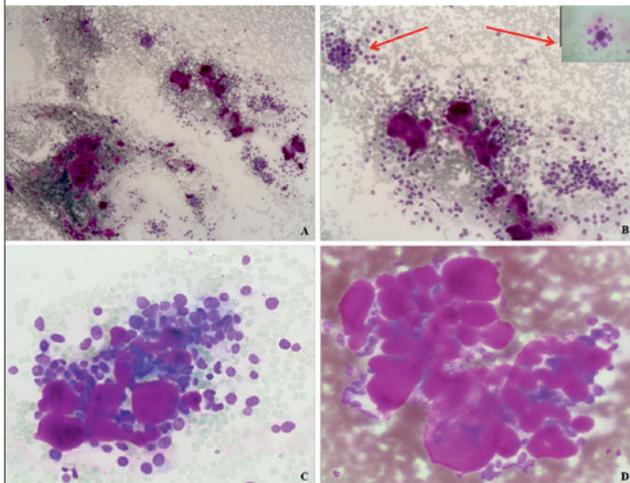
Acknowledgements

The authors would like to thank Prof. Piero Tosi (University of Siena) for his expert review of the manuscript.

Correspondence

Maria Raffaella Ambrosio, Department of Medical Biotechnologies, Section of Pathology, University of Siena, via delle Scotte 6, 53100, Siena, Italy - Tel. +39 0577 585850 - Fax: +39 0577 233235 - E-mail: maradot@libero.it

Fig. 1. A) FNAC revealed highly cellular smears with a monomorphic population of basaloid cells in tight clusters (Figure 1A). B) Partial microfollicle-like pattern. C) The individual cells were round, oval or slightly angulated with fine, granular chromatin, indistinct nucleoli, scant cytoplasm and indistinct borders. D) Small roughly spherical hyaline globules of basement membrane material were found, staining magenta with MGG, and showing marked variability in size.



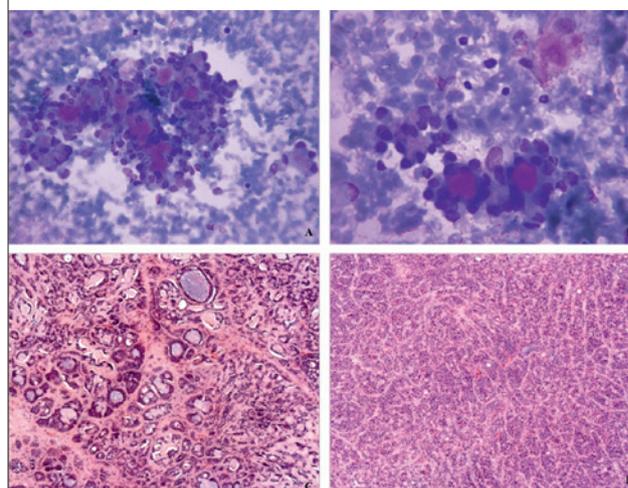
thyroid. Ultrasonography revealed a 16 x 14 mm mixed echogenic lesion in the thyroid parenchyma, involving trachea and soft tissues, without lymphadenopathies. In the suspicion of subacute thyroiditis, fine needle aspiration cytology (FNAC) of the nodule was performed; wet fixed and air dried smears were made and stained with May-Grunwald Giemsa (MGG) and Papanicolaou. FNAC revealed a highly cellular smear with a monomorphic population of basaloid cells in tight clusters (Fig. 1A). There was a partial microfollicle-like pattern (Fig. 1B). The individual cells were round, oval or slightly angulated with fine, granular chromatin and indistinct nucleoli. Cytoplasm was scant, and the cell borders were indistinct (Fig. 1C). There was no definite grooves or intranuclear (pseudo)inclusions. Small hyaline globules of basement membrane material were found. The globules stained magenta with MGG, were roughly spherical and showed marked variability in size (Fig. 1D). "Ropy colloid" was observed. Since an adenoid cystic growth pattern with typical hyaline globules has been reported in tumours other than ACC, differential diagnosis considered numerous entities both primitive (follicular, papillary, anaplastic and medullary carcinoma) and metastatic (mainly from salivary glands and from the laryngotracheal complex). Thyroid primitivity was excluded on the basis of the negativity for TTF1 and thyroglobulin. In addition, computed tomography of neck and thorax did not reveal additional lesions in either major salivary glands or the laryngotracheal complex or lungs. The patient referred an ulcerative lesion of the internal surface of the inferior lip, and thus a FNA was carried out which found the same cytologic features as the thyroid nodule (Fig. 2A-B). The final diagnosis was ACC of minor salivary gland metastatic to thyroid. Diagnosis

was then confirmed by histological examination of the thyroidectomy specimen, which showed an ACC with cribriform and solid pattern (Fig. 2C-D).

Discussion

Thyroid metastases are uncommon, and most originate from the upper and lower respiratory tract, kidney and breast⁹. Primary ACC of the thyroid gland has never been described, and metastases of ACC to the thyroid are rare and mostly originate from the laryngotracheal complex and breast^{4 9 12}. Only one previous case of ACC of major salivary glands (the parotid) metastatic to thyroid has been described, while no metastases of ACC to the minor salivary glands have been observed to date^{6 11 13}. In this report, we describe the first case of minor salivary gland ACC metastatic to the thyroid. ACC represents one of the most malignant tumours of the minor salivary glands¹. It displays unique features, such as slow but aggressive growth, early invasion of peripheral nerves and/or blood vessels and a high incidence of recurrence and distant metastases, mostly in the lungs⁴. The presence of a significant percentage of solid growth, as in our case, implies poor prognosis⁵. It has to be remembered that an ACC-like pattern may also be found in primary malignant tumours of the thyroid, mainly in papillary carcinoma¹⁴⁻¹⁶. However, in these cases, the hyaline globules are early psammoma bodies or thick colloid instead of basement membrane substance of myoepithelial origin. In addition, several cytologic parameters may be helpful in differential diagnosis between thyroid metastases of ACC and primary ACC-like malignant tumours of the thyroid (Tab. I)¹⁷⁻¹⁹. The FNA smears from the thyroid nodule in our case showed all the cytologic features that are diagnostic of

Fig. 2. A-B) FNA of the ulcerative lesion of the internal surface of the inferior lip, showing the same cytologic features as the thyroid nodule. C-D) Histological examination of this lesion showed an ACC with a cribriform and solid pattern metastatic to the thyroid.



Tab. 1. Differential diagnosis between metastatic adenoid cystic carcinoma and primary malignant tumours of the thyroid gland with adenoid cystic pattern.

	ACC	PTC	FTC	MTC	AC
Pattern	cribriform	papillae	microfollicular	discohesive	discohesive
Cytoplasm	basaloid	squamoid	minimal	moderate, with NE granules	variable
Nucleus	oval with coarse chromatin	round with grooves, distinct nucleoli and inclusions	round with fine chromatin	round to spindle, with distinct nucleoli	irregular, with large or multiple nucleoli, coarse chromatin and numerous and atypical mitoses
Other features	hyaline globules	ropy colloid, psammoma bodies	no colloid	amyloid	necrosis

AC: anaplastic carcinoma; ACC: adenoid cystic carcinoma; FTC: follicular thyroid carcinoma; MTC: medullary thyroid carcinoma; NE: neuroendocrine; PTC: papillary thyroid carcinoma.

ACC described in the literature. Moreover, computed tomography of the neck and thorax was negative, and the negativity for TTF1 and thyroglobulin ruled out primary thyroid carcinoma. The present case highlights the need to be aware of possible metastatic thyroid localization of ACC also originating in minor salivary glands of the oral cavity. This is a very rare event, but it should be taken into consideration, and clinical and cytological findings must be carefully examined. In our case, correct diagnosis was made only after an accurate physical examination of the patient which identified an ulcerative lesion of the oral cavity that resulted to be ACC.

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