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CASE REPORT

# Sclerosing mucoepidermoid carcinoma with eosinophilia of the thyroid

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## ABSTRACT

Sclerosing mucoepidermoid carcinoma with eosinophilia (SMECE) is a recently described carcinoma of the thyroid gland associated with Hashimoto's thyroiditis and considered to have a relatively indolent clinical course. We present a 65-year-old female, clinically euthyroid, who presented with a thyroid swelling since last 8 months along with right-sided cervical lymphadenopathy. Fine needle aspiration cytology suggested poorly differentiated carcinoma along with the involvement of the lymphnodes while histopathological examination showed features of SMECE with metastatic deposits in the right-sided cervical lymphnodes.

**KEY WORDS:** Eosinophilia, mucoepidermoid carcinoma, sclerosing, thyroid

## INTRODUCTION

The thyroid gland is unique among endocrine organs in many ways. It is the largest of all endocrine glands, and because of its superficial location, it is the only one that is amenable to direct physical examination and biopsy. Diseases of the thyroid, including a vast array of developmental, inflammatory, hyperplastic, and neoplastic disorders, are exceedingly common in clinical practice.

Thyroid cancer accounts for 0.4% of all cancer deaths with a mortality rate of 8 in 1.5 million people per year. Primary mucoepidermoid carcinomas of the thyroid gland are extremely rare neoplasms. As in the salivary glands and elsewhere in the body, they are composed of admixtures of squamous and mucin-producing glandular cells.

## CASE HISTORY

A 65-year-old female presented with a thyroid swelling on the right side of neck for 8 months. She was euthyroid clinically. On examination, the swelling was $4 \times 4$ cm$^2$, firm, mobile, and moving well with deglutition. There was associated right-sided cervical lymphadenopathy. Her general and systemic examinations were normal. Her thyroid function tests were normal and thyroid scan showed a cold nodule in the right lobe of thyroid. Fine needle aspiration cytology (FNAC) of the thyroid gland along with the right-sided cervical lymphnodes suggested poorly differentiated carcinoma with metastatic deposits in the lymphnodes. A subtotal thyroidectomy was performed. Grossly, the right lobe of thyroid measures $4 \times 2 \times 2$ cm$^3$, and showed a well-circumscribed homogenous grey-white lesion occupying the entire lobe [Figure 1]. The left lobe measures $2 \times 1 \times 1$ cm$^3$, and was grossly unremarkable. The right-sided lymphnode measured $2 \times 1 \times 1$ cm$^3$, with the cut section showing grey-white areas. On histopathological examination (HPE), the right lobe showed a circumscribed nodule consisting of nests of cells with dense sclerosis, intermediate cells with moderate amount of pale eosinophilic cytoplasm, cells in adenoid pattern as well as keratin pearl formation [Figure 2]. There was minimal pleomorphism and no mitotic activity. Stroma showed infiltration by eosinophils, lymphocytes, and plasma cells. No focus of papillary, follicular, or medullary carcinoma was seen in multiple sections studied. Mucicarmine and periodic acid schiff (PAS) with Alcian blue showed focal mucin positivity while Mason's trichrome confirmed the fibrosis. On immunohistochemical studies, the tumor cells showed negativity for thyroglobulin and calcitonin. The left lobe was within normal histological limits.

Multiple sections from the right-sided cervical lymphnodes showed identical features with hematoxylin and eosin (H&E), special stains, and immunohistochemical stains. The final diagnosis of sclerosing mucoepidermoid carcinoma with eosinophilia (SMECE) with metastatic deposits in the right-sided cervical lymphnode was given.

The patient is being regularly followed up in the out patient department (OPD) without any complications.

## DISCUSSION

Primary mucoepidermoid carcinomas in the thyroid gland are extremely rare neoplasms. The first case was described by Rhatigan *et al.*[1] The exact histogenesis remains unknown and controversial. Cytomorphologic descriptions of mucoepidermoid carcinomas of thyroid gland are scant. Many patients with mucoepidermoid carcinomas are women with Hashimoto's thyroiditis.[2] For the most part, mucoepidermoid carcinoma is considered to be a low-grade cancer

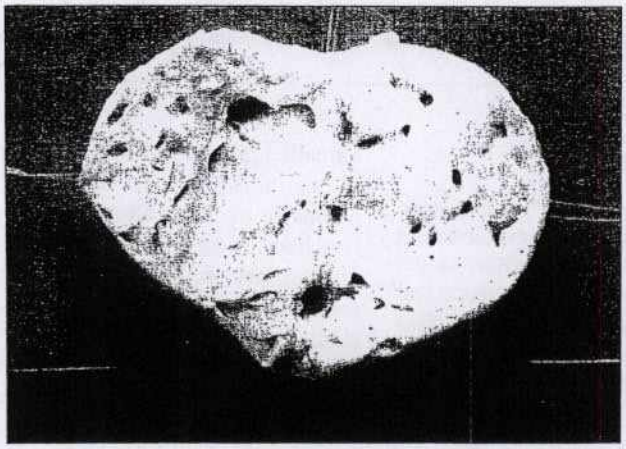

**Figure 1: Gross appearance of the right lobe of thyroid showing a grey-white lesion**

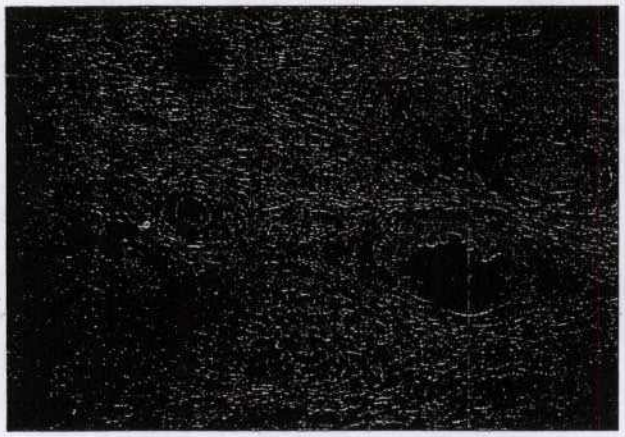

**Figure 2: Photomicrograph showing tumor cells along with a keratin pearl (H&E, x400)**

with a relatively good prognosis. However, examples of distant metastasis have been documented.[2] Several reports have been made of mucoepidermoid carcinoma of the thyroid gland mixed with papillary carcinomas.[3]

Two types of mucoepidermoid carcinomas of thyroid are known to occur: (1) The mucoepidermoid carcinomas of the conventional type (MEC). (2) Sclerosing mucoepidermoid carcinomas of the thyroid gland with eosinophilia (SMECE). The SMECE of thyroid is characterized by unique histologic and biological behavior. The origin of SMECE remains controversial. However recent studies based on immunohistochemistry showed MEC and SMECE differ in their immunostaining properties.[4] MEC is thyroglobulin positive, whereas SMECE is negative for both thyroglobulin and calcitonin. This highlights the fact that SMECE may have originated from the vestiges of the ultimobranchial body.[4] MEC is seen commonly in females (M:F:1:2) and can occur in any age group. Histologically, MEC consist of mucinous cysts of variable sizes along with solid areas of epithelial cells. They have no association with thyroiditis and may occasionally show foci of papillary carcinoma thyroid.

However, SMECE is a distinctive tumor which has extensive fibrosis and eosinophilic infiltration in the stroma. SMECE mainly involves the adult age group (40-75 years) with a female predominance (M:F:1:17).[2] On HPE, identical tumoral features were identified by us and Chan *et al.*[5] and consist of delicate and anastomosing cords and relatively small nests of neoplastic cells situated within a dense hyalinzed stroma. Most of the malignant cells had a squamoid appearance with, prominent nucleoli, intercellular bridges, and keratin pearls. The glandular or mucin-producing component was less conspicuous. Mucinous material was present within lumens and between squamoid cells. Rarely, intracytoplasmic mucin vacuoles were evident. A characteristic feature was the marked infiltration of the tumor cell aggregates and stroma by eosinophils. Similar to us, Chan *et al.*[5] faced difficulty in distinguishing between squamoid tumor cells and metaplastic squamous cells. More recently, Wenig *et al.*[6] described six cases of primary mucoepidermoid carcinoma of the thyroid gland. Like us, all of their patients also had an uncomplicated clinical follow-up period.

Bondeson and Bondeson[7] published a similar case report diagnosed by aspiration biopsy in a 74-year-old woman where the tumor cells were arranged singly and in clusters. In their case, malignant cells were relatively small with spindle-shaped nuclei and high nuclear/cytoplasmic ratios. Chromatin was fine and uniformly distributed; nuclear grooves and pseudoinclusions were occasionally noted. Inflammatory cells, including eosinophils, were present among the tumoral elements.

The differential diagnosis of mucoepidermoid carcinoma of the thyroid gland includes several different entities. Squamous differentiation within otherwise typical papillary adenocarcinomas is a common occurrence.[2] At time squamous differentiation may be extensive giving rise to several diagnostic pitfalls. On the other hand, Chan and Tse[8] have reported mucin production in substantial proportion of metastatic papillary thyroid gland carcinomas. Another consideration is the extremely uncommon "pure" squamous cell carcinoma of the thyroid gland. Many of these cases seem to coexist with anaplastic thyroid carcinoma. However, the degree of anaplasia and pleomorphism in these high-grade malignancies would not be expected in an SMECE.[2] SMECE needs to be also distinguished from medullary carcinomas (with thymus-like differentiation) which will have predominantly plasmacytoid cells, spindle-shaped cells along with extracellular amyloid.[9] Rarely, follicular adenomas and follicular carcinomas may show prominent mucinous differentiation along with a signet ring like appearance.[10] Neither squamous differentiation nor eosinophils should be present in such cases, which will help us to identify SMECE accurately. Finally, neoplasm metastatic to the thyroid gland must also be considered in the differential diagnosis of primary mucoepidermoid carcinomas, including SMECE. In such cases, a through clinical examination including

detailed pathological and radiological information will help us in reaching a final conclusion.

## CONCLUSIONS

Our case report highlights two important features, i.e. although SMECE is considered to be a relatively indolent tumor, occasional metastasis do occur and rarely SMECE may not be associated with Hashimoto's thyroiditis.

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