Conservative Management of Sclerosing Mucoepidermoid Carcinoma with Eosinophilia of the Thyroid Gland: 12 Years Follow-Up after an Atypical Initial Therapy

Keywords: Sclerosing mucoepidermoid carcinoma; Thyroid; Incomplete thyroidectomy

Abstract

Objective: Sclerosing Mucoepidermoid Carcinomas (SMEC) are low-grade malignant tumors with both mucinous and squamous differentiation representing less than 1% of thyroid malignancies. SMEC are sometimes associated with Hashimoto’s thyroiditis and eosinophilia and are more common in females. In rare cases, they have showed a more aggressive behavior with local invasion of the surrounding tissues, trachea, esophagus, or lymph nodes. Distant metastasis to lungs, liver, mediastinum, and bone have also been described. SMEC are usually not responsive to Radioactive Iodine Ablation (RAIA). Despite the generally low risk of recurrence or death, total thyroidectomy and neck dissection is the recommended therapy.

Methods: We report the case of a 41 year old female with a SMEC of the thyroid. Hemithyroidectomy without lymphadenectomy was performed. Pathology showed a nodule with a maximum diameter of 1.5 centimeters contacting the capsule. Tumor cells expressed TTF-1, CEA and CK7. No expression for thyroglobulin, calcitonin, cromogranin or CK20 was observed.

Results: Complete resection of the thyroid was not performed in this case, and the patient was not treated with radioactive iodine postoperatively. After 12 years of follow-up, the patient did not develop residual disease, neither recurrent loco regional nor metastatic disease.

Conclusion: Sclerosing mucoepidermoid carcinoma was treated with hemithyroidectomy, no lymphadenectomy was performed. Despite the size of the nodule and the capsular invasion, the patient had a favourable outcome after a twelve year follow-up. This kind of approach should be taken into account in selected patients.

Case Report

A 41-year-old woman with no remarkable medical history was sent to our endocrinology clinic to evaluate the presence of a thyroid nodule. Physical examination showed a palpable mass in the right lobe of the thyroid. An ultrasound scan was performed, which revealed a tumor with a maximum diameter of 1.5 cm. Thyroid function was normal.

Fine-needle aspiration cytology was performed, which showed the presence of follicular and papillary proliferating cells in a lymph node. A malignant tumor was suspected.

A right hemithyroidectomy was performed on July 2004. The resected thyroid tissue measured 4 x 2 x 2 cm and weighed 6.8 grams, with a tumor measuring 1.5 x 1 x 1 cm, and no capsule around it. Parathyroid glands were preserved. In the operating room, surgeons

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suggested that macroscopically it seemed to be a benign lesion. There were no complications after surgery.

In this case, the tumor contacted the thyroid capsule but did not extend further; it was therefore staged as T1.

The histopathology report showed a sclerosing mucoepidermoid carcinoma in contact with the thyroid capsule. Microscopically, the tumor was composed of numerous nests of squamous cells, glandular structures and papillary formations of solid cells in a sclerotic stroma with numerous eosinophils. Tumor cells presented an irregular nucleus, open nuclear chromatin and moderate eosinophilic cytoplasm in a sclerotic background of chronic inflammatory tissue. The surrounding thyroid tissue showed lymphocytic thyroiditis. The dissected cervical lymph nodes were negative for malignancy. The use of molecular markers revealed positivity to TTF, CEA, and CK7, whereas no expression for thyroglobulin, calcitonin, CK20, chromogranin, or Alcian Blue was observed. The Ki-67 labeling index was not analyzed.

In this case, thyroidectomy was not completed and the patient was not treated postoperatively with radioactive iodine.

During medical reviews in the following years, ultrasound did not reveal tumor recurrence or any suspicious nodes. The patient received suppressive levothyroxine therapy for 8 years and is presently taking 50 mcg of levothyroxine daily to control the underlying hypothyroidism.

Discussion

Total thyroidectomy with central neck dissection is the elective therapy for this type of tumor [7]. It is important to remark that it does not usually respond to Radioactive Iodine Ablation (RAIA), and although other treatment modalities such as external beam radiotherapy, carboplatin and doxorubicin have been used, none of them have been very successful [8].

Recent studies suggest that, as this tumor is usually associated with Hashimoto’s thyroiditis, it probably originates from the metaplastic squamous nests typical of Hashimoto’s thyroiditis histology, although other authors suggest that its origin is in the follicular cells [1].

In addition, some cases of aggressive tumors have been published. Chung et al. reported a case of a 57-year-old woman from Korea, who, two years after total thyroidectomy, underwent radical neck dissection because of a recurrent mass. After radiation therapy, a laryngectomy and esophagectomy was performed due to a recurrent carcinoma in the esophageal wall [9]. These authors also reported an additional case of SMEC with metastasis in regional lymph nodes and esophagus [8]. Another case, reported by Purohit et al. described a 70-year-old woman with kidney metastasis from a SMEC treated with total thyroidectomy, total laryngectomy, and bilateral neck dissection. She also received 150 mci of Radioactive Iodine Ablation [8]. Also, Franssila et al. describe a case of SMEC in which the primary tumor was confined within the thyroid capsule and presenting anaplastic areas invading surrounding structures. This patient died 13 months after diagnosis [10]. Polonia et al. describe a 13-year-old girl with a 2.5 cm nodule in the left lobe of the thyroid that histologically resulted in SMEC. A cervical lymph node metastasis was identified. The patient was treated with radioactive iodine [11].

BRAF mutation is the most common genetic alteration in thyroid cancer, occurring in 45% of sporadic papillary thyroid cancers. It is associated with poor clinic pathological outcomes and is an independent molecular prognostic marker in risk evaluation of thyroid cancer [8]. It would be interesting to know its usefulness for this specific tumor; however, it has not been studied yet.

This rare type of thyroid tumor is unique, with a different pathologic classification and also biologically different from the others. A standardized treatment is needed although a consensus or agreement has not yet been made. Additionally, the treatment of metastatic disease is not defined. It is important to find specific clinical, radiological or histological indicators, or molecular markers that will help us predict prognosis and monitor the illness.

Conclusion

Sclerosing Mucoepidermoid Carcinoma is an infrequent tumor of the thyroid. It shows a female predominance and a relatively benign clinical course, most of the reported cases are low-grade tumors [5], but it can also be very aggressive. Histological studies with molecular markers are needed. Total thyroidectomy and neck dissection is the therapy of choice, based on the irregular outcome of some cases. Despite the initial characteristics of the nodule, our patient had a favorable outcome throughout the 12 years of follow-up after a hemithyroidectomy without lymphadenectomy.

References