

Case Report

An aggressive case of sclerosing mucoepidermoid carcinoma with eosinophilia of thyroid: a case report

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Received: 07 March 2017

Accepted: 20 March 2017

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ABSTRACT

Sclerosing mucoepidermoid thyroid carcinoma with eosinophilia (SMECE) of the thyroid is a rare disease with approximately 50 cases reported worldwide. We describe SMECE of the thyroid affecting a 44 year old female and the first reported case of SMECE of thyroid in New Zealand. This case is of interest as the disease course was particularly aggressive with uncommon immunohistology. Our patient presented with a 4 month history of a painless swelling in her right upper neck. CT revealed a diffusely enlarged thyroid with infiltration of the trachea. A total thyroidectomy, tracheal resection and bilateral neck dissection was performed. Histology showed nests and cords of moderately sized epithelioid cells with prominent nucleoli, with focal glands and tubules. Immunohistology was performed. CK14 and epithelial membrane antigen staining was focally positive. The patient went on to receive chemo-radiotherapy. A CT scan later revealed metastasis to her lungs. She died 7 months from her diagnosis. Our case provides an example of aggressive SMECE of thyroid.

Keywords: Carcinoma, Eosinophilia, Mucoepidermoid, Sclerosing, Thyroid

INTRODUCTION

Sclerosing mucoepidermoid carcinoma with eosinophilia of the thyroid (SMECE) is a rare disease first described by Chan et al in 1991.¹ It is characterised histologically by strands and nests of squamous tumour cells with nuclear polymorphism. Eosinophil infiltration is present in all cases. Immunohistologically the tumour cells are positive for cytokeratin and negative for thyroglobulin, and calcitonin. The tumours occur largely in females of middle age (32-73) and all have a history of Hashimoto's thyroiditis.² The tumour has up, until recently, been considered a low-grade malignancy however emerging evidence now suggests it is more aggressive.³

CASE REPORT

A 44 year old Caucasian female presented with a 4 month history of a painless enlarging mass in the left side of the

neck, recent difficulty breathing on exertion, and hoarse voice. She had a 20 year history of hypothyroidism, controlled with 200mcg Thyroxine daily. Examination revealed a diffusely enlarged, hard, thyroid gland adherent to the larynx and trachea. Palpation of her neck revealed a right 3cm level II lymph node and left 1.5cm level III lymph node. Flexible nasendoscopy showed a narrowed subglottis. Vocal cord movement was symmetrical. A fine needle aspiration (FNA) of the thyroid and right level III lymph node was consistent with adenocarcinoma. A CT scan of the chest and neck demonstrated a diffusely enlarged thyroid gland, the left lobe was asymmetrically enlarged, wrapping around the trachea to contact the oesophagus. The left tracheal wall was contiguous with the thyroid, with tracheal invasion suspected. A 21x21mm right level II node and 14x18mm left level III node were identified.

The patient proceeded to operative intervention. A tracheoscopy performed at the beginning of the procedure

revealed invasion of tumour bilaterally into the trachea. An extended total thyroidectomy, tracheal resection with reconstruction, and bilateral cervical neck dissections of levels II, III, IV and VI was performed. At operation, the tumour was close to, but not involving the right cricothyroid joint. The right recurrent laryngeal nerve was thickened, however was functioning on nerve stimulation. 3cm proximal to the cricothyroid joint, the left recurrent laryngeal nerve was surrounded in tumour and was sacrificed. The tumour was closely adherent to the cricoid cartilage and was partially resected. The trachea was then separated from the oesophagus and 4cm of trachea was resected en bloc immediately below the cricoid cartilage. Both cricothyroid joints were preserved. Frozen sections were taken and margins were deemed histologically clear.

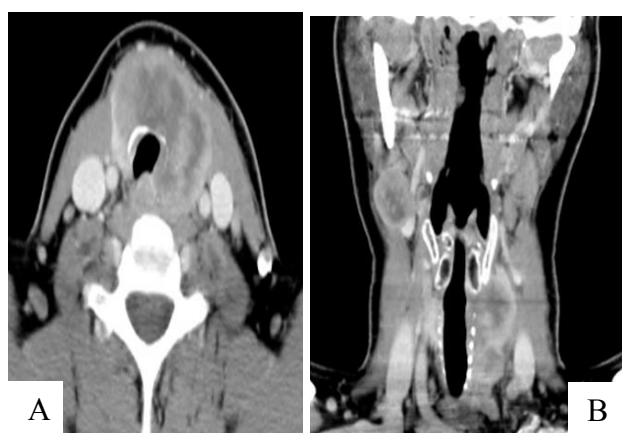


Figure 1: Initial Head and Neck CT. A: Axial view of Thyroid tumour at level of trachea. B: Coronal view of Neck revealing tumour contiguous with trachea and an enlarged right 21x21mm level II node.

The thyroid tumour mass measured 60x40x35mm, with no normal thyroid tissue. Carcinoma was evident in peritracheal soft tissue and there was evidence of transmural invasion of the trachea from superior to inferior margins. The right neck dissection had 5/20 nodes positive, the left had 4/27 nodes positive.

Microscopically there were nests and cords of moderately sized epithelioid cells with prominent nucleoli, with focal glands and tubules. Lymphovascular and perineural invasion was present. Keratinization was not present. CK14 and epithelial membrane antigen (EMA) immunostaining was focally positive. CK7 was mainly positive. Thyroglobulin, thyroid transcription factor 1 (TTF1), carcinoembryonic antigen (CEA), and calcitonin were negative.

Our patient was discharged 9 days after the procedure. Calcitriol and Calcium were prescribed to control hypocalcemia. Two months after the procedure she was admitted for dyspnoea and stridor. Microlaryngoscopy revealed polypoid granulation tissue in the anterior upper trachea. A biopsy of this tissue revealed tumour

recurrence. She commenced radiotherapy shortly thereafter. A CT performed 7 months after her diagnosis revealed disease progression with 8 nodules in the lungs consistent with metastasis. She died days later at home of haemorrhage into her airway.

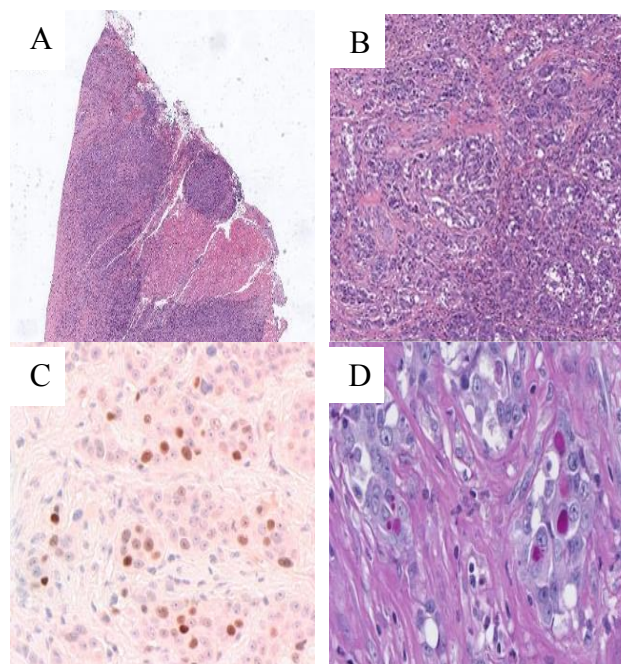


Figure 2: A: Low power magnification showing tumour invading skeletal muscle. B: Medium power magnification showing cords and nests of epithelioid cells in sclerotic stroma. C: Cells staining focally for p63. D: High power magnification PASD Mucin positive cells amongst moderately sized epithelioid cells with prominent nucleoli.

DISCUSSION

This is the first case of SMECE of the thyroid reported in New Zealand. The course of our patient's disease was aggressive and involved distant metastases. Up until recently, SMECE of the thyroid has been considered a low grade malignancy and distant metastases were thought to be uncommon, with some exceptions.⁴⁻⁶ Previous case reports have shown largely good outcomes for patients.⁷ However a recent study by Quiroga-Garza G et al suggests that SMECE of thyroid is more aggressive than previously thought.³ They found distant metastases in 50% of their patients with SMECE of the thyroid. Further, a literature review in 2004 by Shehadeh et al found 4 out of 23 patients with SMECE of thyroid also had distant metastatic disease.²

Our experience, and that of previously aforementioned published cases, raise concern that SMECE may not follow the natural history of a low grade malignancy. In the literature patients are often reported to survive many years after their diagnosis. Our patient deteriorated rapidly and died 7 months from initial diagnosis.

Our patients tumour differed immunohistologically from the majority of thyroid SMECE in that it was not positive for CEA. Shehadeh et al found that 13 of 16 SMECE patients were positive for CEA.² The immunohistological profile of her tumour was otherwise in keeping with the majority of reported cases.

This case provides a further example of particularly aggressive SMECE of the thyroid. To our knowledge there is no patient that has progressed this rapidly from the time of their diagnosis. We hope this paper highlights the emerging evidence of the potentially aggressive nature of SMECE and as per Quiroga-Garza et al facilitates early diagnosis and timely patient follow up to improve patient outcomes.³

Funding: No funding sources

Conflict of interest: None declared

Ethical approval: Not required

REFERENCES

1. Chan JK, Albores-Saavedra J, Battifora H, Carcangiu ML, Rosai J. Sclerosing mucoepidermoid thyroid carcinoma with eosinophilia. A distinctive low-grade malignancy arising from the metaplastic follicles of Hashimoto's thyroiditis. *Am J Surg Pathol*. 1999;15(5):438-48.
2. Shehadeh NJ, Vernick J, Lonardo F, Madan SK, Jacobs JR, Yoo GH, et al. Sclerosing mucoepidermoid carcinoma with eosinophilia of the thyroid: a case report and review of the literature. *Am J Otolaryngol*. 2004;25(1):48-53.
3. Quiroga-Garza G, Lee JH, El-Naggar A, Black JO, Amrikachi M, Zhai QJ, et al. Sclerosing mucoepidermoid carcinoma with eosinophilia of the thyroid: more aggressive than previously reported. *Hum Pathol*. 2015;46(5):725-31.
4. Sim SJ, Ro JY, Ordonez NG, Cleary KR, Ayala AG. Sclerosing mucoepidermoid carcinoma with eosinophilia of the thyroid: report of two patients, one with distant metastasis, and review of the literature. *Hum Pathol*. 1997;28(9):1091-6.
5. Geisinger KR, Steffee CH, McGee RS, Woodruff RD, Buss DH. The cytomorphologic features of sclerosing mucoepidermoid carcinoma of the thyroid gland with eosinophilia. *Am J Clin Pathol*. 1998;109:294-301.
6. Chung J, Lee SK, Gong G, Kang DY, Park JH, Kim SB, et al. Sclerosing Mucoepidermoid Carcinoma with eosinophilia of the thyroid glands: a case report with clinical manifestation of recurrent neck mass. *J Korean Med Sci*. 1999;14(3):338-41.
7. Baloch ZW, Solomon AC, LiVolsi VA. Primary mucoepidermoid carcinoma and Sclerosing mucoepidermoid carcinoma with eosinophilia of the thyroid gland: a report of nine cases. *Mod Pathol*. 2000;13(7):802-7.

Cite this article as: Eason JR, Allison R. An aggressive case of sclerosing mucoepidermoid carcinoma with eosinophilia of thyroid: a case report. *Int Surg J* 2017;4:1780-2.