Hyalinising trabecular adenoma of the thyroid

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ABSTRACT
Hyalinising trabecular adenoma (HTA) of the thyroid is rare and controversies remain regarding the diagnosis and management. It is an uncommon benign thyroid tumour that can present as a solitary thyroid nodule, a prominent nodule in a multinodular goitre, or as an incidental finding in a thyroidectomy specimen. Some considered HTA a unique entity, while others have proved it to be a variant of papillary carcinoma or have considered it a nonspecific pattern that may be seen with a variety of thyroid lesions. We report the case of a 31-year-old Chinese lady who presented with four years history of right sided goitre that was gradually increasing in size. Fine needle aspiration cytology showed appearances of follicular adenoma. Intra-operative frozen section histology of the excised right thyroid lobe was reported as trabecular adenoma which was confirmed with histopathological examination.

Keywords: Thyroid neoplasm, hemithyroidectomy, trabecular adenoma

INTRODUCTION
Hyalinising trabecular adenoma (HTA) of the thyroid gland is a rare, benign neoplasm, first described by Carney et al in 1987. In the recent World Health Organisation classification of tumours of endocrine organs, the term adenoma was replaced by tumour and defined as a rare tumour of follicular cell origin with a trabecular pattern of growth and marked intratrabecular hyalinisation. Debates and controversies concerning this ambiguous neoplasm lie on its cytological identification, classification and management.

CASE REPORT
A 31-year-old Chinese lady was referred to the department with a four years history of right sided goitre that was gradually increasing in size. There were no other associated symptoms. The patient had no past medical history of note and no significant family history of thyroid disease.

The clinical examination revealed a prominent right sided neck swelling which moved on swallowing. On palpation, the neck swelling was firm with normal overlying skin. There were no palpable neck lymph nodes. She was in euthyroid state and tested negative for thyroid antibodies. Fibre-optic laryngoscopy showed normally functioning vocal cords.

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cords. A routine thyroid work-up was performed that included a Doppler ultrasound and ultrasound-guided fine needle aspiration cytology (FNAC). The ultrasound scan of the neck revealed a solid right nodule measuring 3.0 x 3.5 cm. There was also a cystic right thyroid nodule seen lying inferior to the former nodule. Routine baseline blood tests and thyroid function test were within normal limits.

Histological analysis was reported as follicular adenoma, although well differentiated follicular carcinoma could not be excluded. A computed tomography (CT) scan of the neck was performed. The diagnosis of right sided multi-nodular goitre was made with no cervical lymphadenopathy (Figure 1). The patient was scheduled for right hemithyroidectomy with intraoperative frozen section histology and to proceed to total thyroidectomy if the lesion was malignant.

Intraoperative frozen section histology showed trabecular adenoma with no malignant cells. The post-operative recovery was uneventful. The final histopathological report of the excised right thyroid lobe was “compact acini arranged in trabeculated pattern and a focus of papillary structures formed of fibrivascular core and also cuboid cells containing vesicular nuclei. There are nuclear groove and pseudo-inclusions. Although the features are suggestive of trabeculated adenoma (Figure 2), this lesion should be considered as follicular variant of papillary carcinoma”.

Following consultations with the patient, Senior Pathologist and Clinical Medical Oncologist, it was decided that a follow up left completion thyroidectomy be performed. In our practice we treat papillary carcinoma with total thyroidectomy and post-operative radioactive iodine therapy and do follow-up with thyroglobulin levels.

A CT scan of the neck, thorax, abdomen and pelvis performed pre-operatively revealed normal study. A left completion thyroidectomy was done five months after the initial operation. Post-operative recovery was uneventful and she was discharged on the sixth post-operative day.

Fig. 1: A contrast enhanced axial computed tomography scan showing a right sided multinodular goitre (arrow).
Histopathological examination of the resected specimen was reported as normal thyroid tissue with no malignant cells seen. As discussed with the Clinical Medical Oncologist, radioactive iodine therapy was not required postoperatively as there were no malignant cells. At the last follow up eight months following surgery, the patient remains well and her serum thyroglobulin level remains very low.

DISCUSSION

HTA is an uncommon, benign thyroid tumour. Hyalinising trabecular neoplasm of the thyroid was originally described in 1987 by Carney et al. as "uncommon but potentially confusing neoplasm". Carney studied 119 neoplasms of this type, collected over 20 years (1987-2007) and found them to be mostly benign. However, one tumour had vascular and capsular invasion and pulmonary metastasis. We present our first recorded case of HTA in Brunei Darussalam.

HTA can present as a solitary thyroid nodule, a prominent nodule in a multinodular goitre or as an incidental finding in a thyroid-decortomy specimen. The tumour consists of polygonal, oval and spindle cells arranged in a trabecular pattern and separated by a hyalinised stroma. A uniform and diffuse solid and trabecular architecture is the hallmark of HTA. There is controversial surrounding this lesion as some pathologists and endocrinologists have considered it a unique entity, while others have proved it a variant of papillary carcinoma and still others have considered it a nonspecific pattern that may be seen with a variety of thyroid lesions. It is confusing because there are other thyroid neoplasms that show focal areas of trabecular pattern, therefore fine needle aspiration cytology analysis is not straightforward. HTA is frequently misdiagnosed as papillary carcinoma, medullary thyroid carcinoma or primary thyroid paragangliomas on FNAC. It may be misinterpreted as the encapsulated variant of medullary carcinoma due to the presence of elongated spindle cells and a stromal hyaline fibrosis that mimics amyloid. This can be distinguished by positive thyroglobulin immunoreactivity and negative calcitonin immunoreactivity. HTA may also exhibit features of paraganglioma which include a nested Zellballen architecture coinciding with a delicate vascular network. However, unlike HTA, paragangliomas are non-reactive for thyroglobulin and reactive for neurofilaments.
The main dilemma is in the differentiation between papillary thyroid carcinoma and hyalinising trabecular neoplasm (HTN) as they share similar histopathological characteristics such as nuclear grooves, pseudo-inclusions and psammoma bodies and RET proto-oncogene rearrangements. Areas with hyalinising trabecular pattern have been identified within both primary and metastatic papillary thyroid carcinoma. At present, there are ongoing controversies with regards to its classification as a variant of papillary carcinoma or as a separate entity and on its management which varies from total thyroidectomy with radioactive iodine therapy, total thyroidectomy alone or thyroid lobectomy. Zhu et al. studied ways to differentiate between papillary thyroid carcinoma and HTN with immunohistochemical tests such as CK-19, a type of acid cytokeratin, Galectin-3, a galactosid-binding protein and HBME-1. These thyroid malignant tumour markers are intensively expressed in papillary thyroid carcinoma but only weakly in benign lesions. In future immunohistochemistry maybe useful to differentiate between follicular variant of papillary carcinoma with prominent hyalinisation and trabeculations. However, this require further studies. Another controversial aspect of HTA is that it can co-exist with papillary carcinoma.

Recent literatures have emphasised the fact that HTA can be malignant with vascular and capsular invasion and metastatic potential. With this in mind, the tumours are now referred as hyalinising trabecular tumour (HTT) rather than HTA.

In conclusion, although HTT is rare, clinicians need to be aware of this entity as controversy and confusion still surround this entity. Some believe that HTA may represent a malignant neoplasm of low metastatic potential while others considered it a variant of papillary thyroid carcinoma.

REFERENCES