CASE REPORT

De novo papillary carcinoma in a thyroglossal duct cyst

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Introduction

Thyroglossal duct cysts (TGDC) account for 70% of congenital neck masses1. Majority (70%) present during childhood and 7% in adulthood2. Only 1% of thyroid carcinomas develop in a TGDC3. A TGDC presents as an enlarging painless anterior neck mass in children or young adults, and is usually a clinical diagnosis. It is essentially impossible to clinically differentiate thyroglossal duct carcinoma from benign TGDC. A preoperative diagnosis may be suggested by ultrasound guided fine needle aspiration cytology. It is essential to differentiate primary papillary carcinoma in a TGDC from a metastatic papillary carcinoma of thyroid.

Case presentation

A 23 year old girl presented with a painless lump over the anterior neck for one month. The lump was gradually enlarging in size with no recent rapid enlargement. She had no local pressure symptoms such as difficulty in breathing or swallowing. There were no symptoms suggestive of hyper or hypothyroidism. Her past medical history was unremarkable with no adverse drug reactions or allergies. There was no family history of thyroid malignancies.

She was not pale or icteric. There was a non-tender firm midline lump located just above the hyoid bone. It was 2cm in diameter, spherical and smooth with well-defined edges. It was fluctuant but not transilluminable. Lump moved up with swallowing and also on protrusion of the tongue while stabilizing the jaw. Overlying skin was normal. No cervical lymphadenopathy or palpable thyroid nodules. The base of the tongue showed no ectopic thyroid tissue. No features of hyper or hypothyroidism. The other systems were normal.

Diagnosis and management

Ultrasound (USS) of the neck revealed a suprahyoid thyroglossal cyst and a sub-centimetre solitary thyroid nodule (STN) in the right lobe with no cervical lymphadenopathy. FNAC of the TGDC showed a benign cystic neoplasm. US guided FNAC of the right thyroid nodule was performed to exclude a primary papillary carcinoma of the thyroid which only revealed benign cells. Thyroid functions were normal.

A Sistrunk operation was done. Histology of the TGDC revealed a papillary carcinoma confirmed by immunohisto-chemical studies as CTF1 nuclear and CK19 positivity in cells lining the papillae.

A Total Thyroidectomy was performed following an oncological consultation to obtain a definitive tissue diagnosis of the thyroid nodule and to facilitate adjuvant treatment.

Histology of the thyroid nodule was of a papillomatous focus likely to be hyperplastic and rest of the gland normal. Immunostains were done on the suspicious focus and CK19 was negative confirming a benign lesion. Patient is currently on Thyroxine and is regularly followed up at our clinic and the oncology clinic.

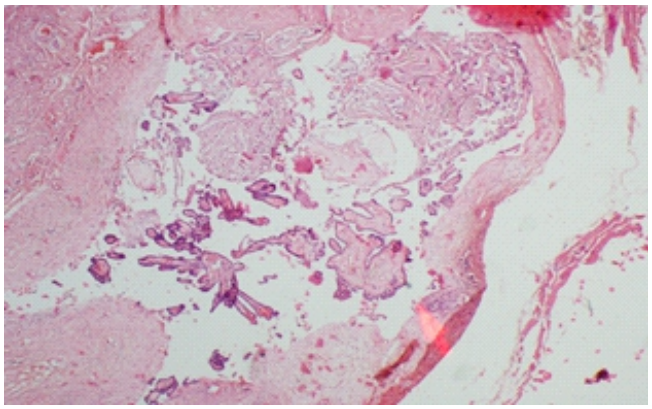

Figure 1. H&E Section of TGDC showing papillary proliferation with minimal nuclear changes suspicious of papillary carcinoma.

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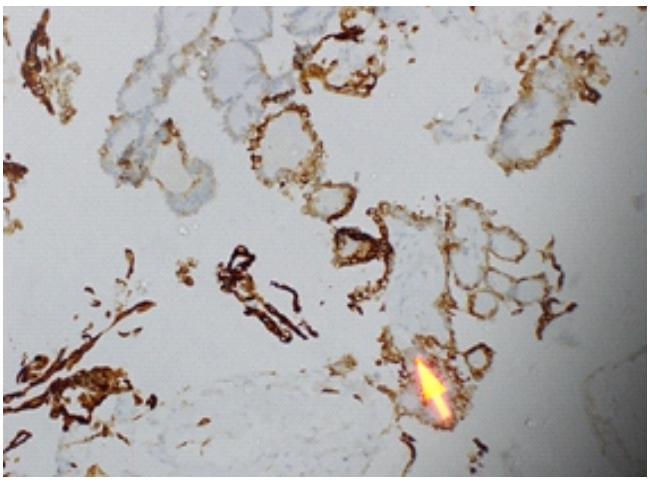

Figure 2. Immunohistochemical section of TGDC showing CTF1 nuclear positivity & CK19 diffuse membrane positivity in the cells lining the papillae confirming papillary carcinoma arising in a TGDC.

Discussion

TGDCs are the commonest developmental anomaly of the thyroid. However TGD carcinomas are very rare, with the majority arising from thyroid remnants4. Papillary carcinoma is the most common type (94%) with less than 5% squamous cell in origin5. The aetiology is uncertain. Two theories exist to explain the origin of TGD carcinomas. One is the de novo theory of origin of papillary carcinoma from a TGDC where the thyroid will be normal as in this case study. Secondly the metastatic spread from an occult primary papillary carcinoma of the thyroid due to its multifocal nature. Multifocal growth is common in papillary thyroglossal carcinomas, with a second lesion present in 10% of cases. Total thyroidectomy should be considered for differentiated thyroid malignancy in a thyroglossal cyst. Conservative management is only advocated in females less than 40 years of age, no capsular invasion and tumour size less than 1cm. Large tumours require total thyroidectomy followed by radioiodine ablation plus TSH suppression as for a thyroid malignancy. In the presence of positive cervical lymph nodes a cervical block dissection is indicated. On the other hand no additional treatment is needed apart from Sistrunk procedure for squamous cell origin of TGD carcinoma.

Conclusion

De novo papillary carcinoma in a TGDC is rare. Often it is an incidental diagnosis after surgical resection. Total thyroidectomy would be indicated in some patients.

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Learning Points:

- Surgically excised Thyroglossal duct cysts should always be sent for histopathology.
- It is essential to differentiate primary papillary carcinoma in a TGDC from a metastatic papillary carcinoma of thyroid.