

## PAPILLARY ADENOCARCINOMA OF THYROID IN A PATIENT WITH RIGHT SUBMANDIBULAR MASS - A RARE CASE OF 'LATERAL ABERRANT THYROID'

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### ABSTRACT

**Background:** Ectopic thyroid is a rare entity in the study of thyroid disease. The occurrence of ectopic thyroid tissue as a mass in the submandibular region is even rarer.

**Aim:** To report a case of papillary adenocarcinoma of thyroid within a right submandibular mass in a 67 year-old man. Additionally this is to alert doctors on the possibility of the condition and re-emphasize the need for proper and thorough histologic investigation.

**Method:** The case notes of the patient, who had presented a year earlier with right submandibular mass, were retrieved and studied. At the initial presentation chest radiograph and assessment of the oropharynx by an otorhinolaryngologist were carried out. Full blood count, erythrocyte sedimentation rate (ESR) and fine needle aspiration biopsy (FNAB) were done at the second presentation. An ultrasound scan of the neck and repeat chest radiograph were also performed.

**Results :** The right submandibular mass had increased in size about 10-fold by the time the patient was seen 13 months later. The chest radiograph and oropharyngeal examination were normal. The neck ultrasound scan demonstrated the presence of a solid right submandibular mass. The FNAB showed papillary adenocarcinoma of the thyroid. He declined a near-total thyroidectomy and demanded nonsurgical treatment. Low dose L-thyroxine (0.1mg) daily was started and referred to another centre for possible radioactive iodine treatment. This latter treatment could not be given because of lack of facilities. Follow-up review in 3 months showed that the patient was only on L-thyroxine therapy and his clinical condition had deteriorated. He was subsequently lost to follow-up.

**Conclusion:** This patient illustrates the even rarer case of a 'lateral aberrant thyroid' presenting as a malignant submandibular mass. It is probably the latest addition to the rather rare number of such cases in the literature. Doctors should be aware of the possibility of the condition and the need for proper and thorough histologic investigation is re-emphasized.

**Keywords:** Submandibular mass, aberrant thyroid, papillary adenocarcinoma.

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### INTRODUCTION

It has been stated that there is no evidence that aberrant thyroid tissue ever occurs in a lateral position<sup>1</sup>. The presence of 'normal thyroid tissue' in such a position, and separate from the thyroid gland should be considered and treated as a metastasis in a cervical lymph node from an occult papillary thyroid carcinoma<sup>1</sup>. It is therefore a widely held belief that the so called 'lateral aberrant thyroid' represents metastatic thyroid cancer<sup>1-6</sup>. The condition is not common and it is even rarer to find ectopic thyroid tissue presenting as a submandibular mass<sup>7</sup>. It is now known, however, that not all 'lateral aberrant thyroid' tissue is malignant<sup>2,3,6</sup> as cases benign thyroid tissue with normal function as well as those with hyperfunction have been described<sup>3,4</sup>. A case of

papillary carcinoma of an aberrant thyroid tissue presenting as a right submandibular mass is reported and could be the latest addition to the rather rare number of such cases reported in the literature.

**CASE REPORT:** A 67-year-old man was first seen in 2004 with a 4-month history of a swelling in the submandibular region. The swelling was painless and patient denied any oral pathology or cough. Clinical examination confirmed a nodular mass in the right submandibular area. There were no other cervical swellings. The oral cavity was normal with a healthy dentition. Chest and abdominal examination, as well as oropharyngeal assessment by the otorhinolaryngologist proved normal. Hemoglobin level was 12.1g/dl with normal white cell count and differentials. The ESR was not raised and chest radiograph was normal. An excisional biopsy of the mass was advised but patient refused surgery. He reported back in 2005, thirteen months after the first

presentation. The right submandibular mass had increased in size about 10-fold and had become painful. A repeat chest radiograph was still normal. The ESR was raised on this occasion while the hemoglobin had dropped to 10.7g/dl. White cell and differential counts were still within normal range. FNAB was done and the report showed papillary carcinoma of the thyroid. On receipt of the histopathological report, T<sub>3</sub> T<sub>4</sub> TSH levels were done and found to be (normal). An ultrasound scan of the neck on this occasion showed the thyroid gland to be normal both in size and position. FNAB was not due on the normal gland. Patient was offered total thyroidectomy but refused and requested treatment with drugs. At his age he did not think he could survive such major surgery. He was started on low dose L-thyroxine (0.1mg daily) and referred to another centre for possible radioactive iodine treatment. Unfortunately at review 3months later, he had not had the radioactive iodine therapy because of lack of facilities. The L-thyroxine treatment was continued despite his deteriorated clinical condition. He could not make the next 3-month review appointment and was subsequently lost to follow-up. Unfortunately, it was not possible to trace him because of logistics and financial constraints of the medical social workers unit.

## DISCUSSION

The presence of the aberrant thyroid tissue is a rare entity in the study of thyroid disease<sup>8</sup>. This so-called 'lateral aberrant thyroid' tissue had been regarded as representing a metastasis from a malignant tumor of the thyroid gland<sup>2</sup>. This gave rise to the surgical dictum that the so-called 'lateral aberrant thyroid' represents metastatic thyroid cancer<sup>3</sup>. It is now known that not all 'lateral aberrant thyroid' is malignant<sup>3</sup>. Kozol *et al* reported 16 cases of patients with benign ectopic thyroid tissue<sup>3</sup>. Seven of these cases were discovered during evaluation and treatment of hyperparathyroidism and the remaining 9 cases diagnosed during the management of thyroid disorders or cervical nodules<sup>3</sup>. These authors have therefore suggested that the old dictum stating that lateral aberrant thyroid represents metastatic cancer should be either modified or removed in review texts and surgical examinations<sup>3</sup>. The occurrence of ectopic thyroid tissue as a mass in the submandibular region is even rarer. As at 1996 only three such cases had been reported<sup>7</sup>; by 2001 the number had increased to eight<sup>5</sup>. The diagnosis is made by the use of imaging techniques like ecography, computerized tomography, ultrasonography and radionuclide scintigraphy<sup>5,7,9</sup>. Excision biopsy or fine needle aspiration biopsy<sup>9</sup>, (as was done in this case report) gives the histologic diagnosis. In the report by DeLong *et al* excisional biopsy was made possible in 40

Out of 52 patients<sup>9</sup>. This mode of biopsy was advised when the patient was first seen in 2004 with a small swelling. Once the diagnosis of papillary adenocarcinoma has been made the treatment is total thyroidectomy. Unilateral or bilateral modified neck dissection is done depending on the extent of cervical lymphadenopathy<sup>9</sup>. If a near-total thyroidectomy is offered, a post operative radioactive iodine therapy is advised. Such treatment is designed to eradicate any remnant thyroid tissue. Surgical treatment was refused in this case and only L-thyroxine could be offered because of lack of facilities for radioactive iodine treatment.

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