

Papillary Carcinoma arising in the Lingual Thyroid: A Case Report and Review of the Literature

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Abstract

The lingual thyroid is an established developmental abnormality caused by a failure of descent of the thyroid gland or remnants, from the foramen cecum to the pretracheal area. Malignant transformation of ectopic lingual thyroid is rare with just 41 cases reported in the literature. Only 8 of these cases were papillary carcinoma. We report a case of papillary carcinoma of the lingual thyroid along with a review of the literature.

INTRODUCTION

The thyroid gland develops from a first pharyngeal derivative as a median diverticulum in the floor of the pharynx during the fourth embryonic week. The diverticulum located at the foramen cecum migrates caudally to lie anterior to the developing trachea. The gland is attached to the foramen cecum by the thyroglossal duct. Atrophy of the thyroglossal duct occurs from the seventh embryological week. Complete failure of descent of the gland from the foramen cecum leads to the formation of a lingual thyroid.

Symptomatic lingual thyroid tissue is unusual with approximately 400 previously reported cases (1). Carcinoma of the lingual thyroid is a rare clinical entity with an estimated incidence of 1% (2). The rate of malignant transformation in ectopic thyroid tissue is no greater than in the normally placed thyroid gland (3). Follicular carcinoma of the lingual thyroid is the commonest histopathological subtype (4). There is currently no explanation for the apparent follicular carcinoma predominance in cancers of the lingual thyroid which contrasts with other carcinomas of ectopic thyroid tissue where papillary carcinoma dominates (4, 5). Only eight cases of papillary carcinoma of the lingual thyroid have previously been reported.

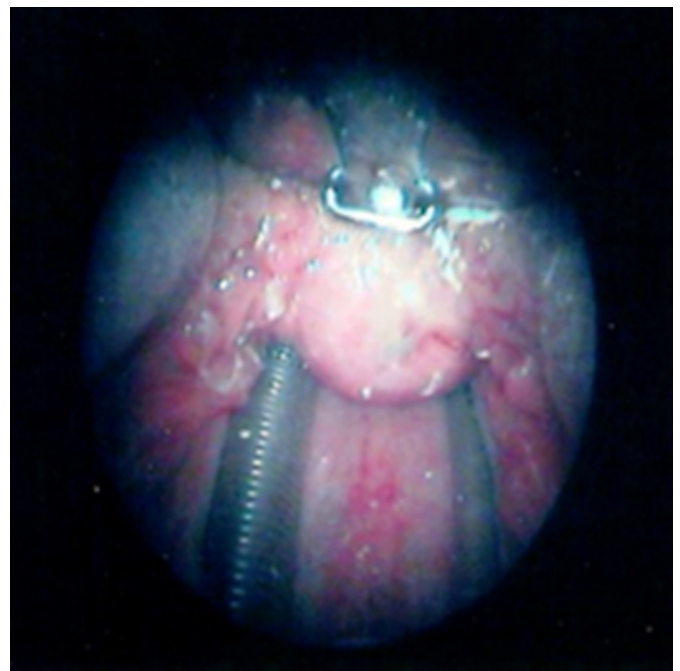
CASE REPORT

A 50 year old lady presented with a five year history of a croaky voice and a sensation of a foreign body in the throat on swallowing. Fibre optic nasendoscopy revealed a mass arising from the posterior third of the tongue. A direct

laryngoscopy was performed to reveal a 2 x 2 cm rounded mass on the base of tongue that was firm in consistency (figure 1).

Figure 1

Figure 1: Lingual Thyroid appearance on direct laryngoscopy



When a biopsy was taken, the centre was found to be necrotic. Follicular thyroid tissue was found at biopsy consistent with a diagnosis of lingual thyroid. However, the histopathologist was not completely able to exclude the presence of a well differentiated follicular carcinoma due to insufficient capsule taken at biopsy.

A Computerised Tomography (CT) scan of the neck with contrast demonstrated a 2 x 2 cm lesion in the midline on the posterior aspect of the tongue with containing scattered areas of calcification (figure 2). The lesion reached inferiorly to the level of the hyoid bone. Very little thyroid tissue was present in the neck.

Figure 2

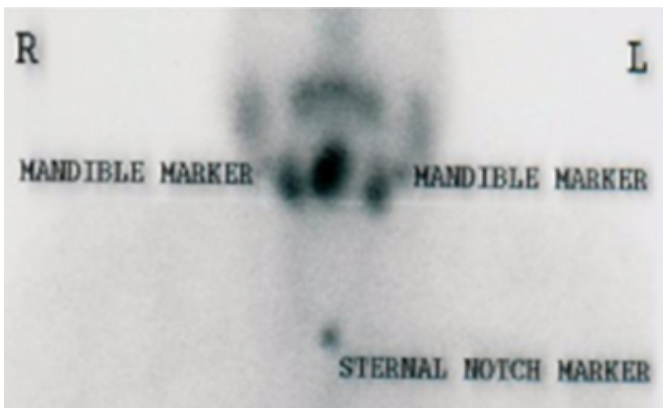
Figure 2: CT scan demonstrating the tongue base lesion



A further isotope thyroid scan was performed to reveal uptake between the salivary gland and nasopharynx indicating a lingual thyroid. No activity was seen in the thyroid bed (figure 3). Thyroid function tests were normal.

Figure 3

Figure 3: Isotope thyroid scan demonstrating the lingual thyroid

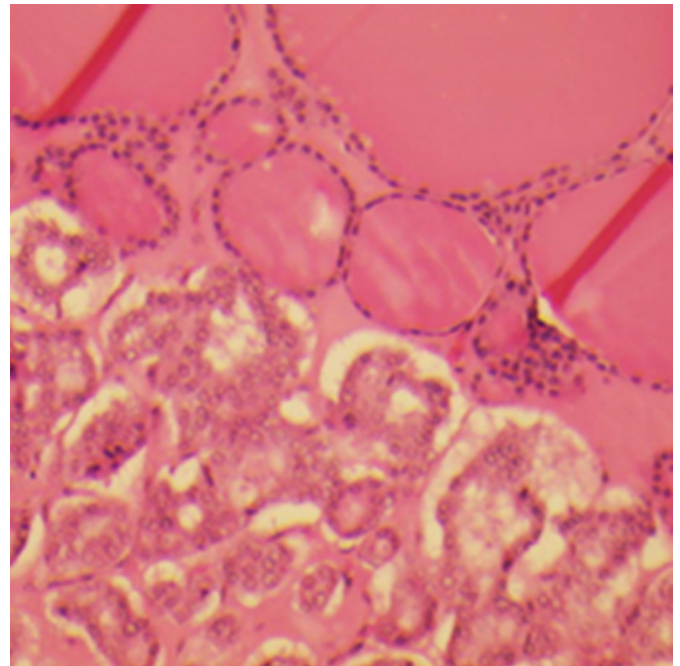


Surgery was performed to remove the mass as the previous histology was inconclusive. The lingual thyroid was

removed via supra hyoid pharyngotomy. A temporary tracheostomy was performed. The histology was reported as well differentiated follicular variant papillary carcinoma of the thyroid measuring 4 x 3.5 cm (Figure 4). The nodule was only 3 mm from the deep margin.

Figure 4

Figure 4: Histology picture of papillary carcinoma of lingual thyroid



The patient made a good recovery following surgery. Post operatively she was commenced on 150 mcg thyroxine. Two months following surgery she underwent radio-iodine ablation (3,500 MBq of I-131). The I-131 tracer test showed only background uptake six months later. Two years following surgery she remains well with the thyroglobulin level being unrecordable and TSH < 0.01 mU/litre.

DISCUSSION

Symptomatic lesions of the lingual thyroid may appear at any time from birth to old age (3). There is a slight female predominance similar to that found in carcinoma of normally placed thyroid tissue in which an approximate 2:1 female-to-male ratio exists (4). Symptoms are largely because of mass effect and include dysphonia, dyspnoea, haemoptysis, or a foreign body sensation (3,4). The majority of lingual thyroids arise during puberty, pregnancy or menses with an average age of 40 years (3).

Examination findings include the presence of a midline lingual mass and often the absence of a palpable pretracheal thyroid. The differential diagnosis includes thyroglossal

cyst, lipoma, lymphangioma, angioma, fibroma, lymphoma, dermoid cyst, squamous cell carcinoma, mucous retention cyst and hypertrophied lingual tonsil tissue (2, 6).

Between two thirds and three quarters of patients with symptomatic lingual thyroids will have no other functioning thyroid tissue and approximately 70% of patients will be hypothyroid (3). Our patient was found to be euthyroid at presentation but radionucleotide scans confirmed the absence of thyroid tissue in the neck. Computerised tomography can be useful in defining the size of the lesion. Radionucleotide technetium 99m and iodine 131 or 123 thyroid scans are important to confirm the presence of ectopic thyroid tissue within the tongue and to assess for thyroid tissue within the neck. Clinically and radiographically lingual thyroid carcinoma presents in an identical fashion with that of a symptomatic benign lingual thyroid mandating the need for biopsy or aspiration (4).

Malignancies arising from the lingual thyroid can be of varying histopathological types (7) (Table 1).

Figure 5

Table 1: Showing histopathological types of the reported cases of malignancies of lingual thyroid (with references).

Histopathological Type	Number
Follicular carcinoma (3,7, 15-21)	11
Papillary carcinoma (4-5,12-13,22-23)	8
Adenocarcinoma (24-28)	5
Hurthles cell carcinoma (29-30)	2
Undifferentiated (31)	1
Unclassified(32-43)	14
Total	41

Follicular carcinomas have been seen more frequently than papillary carcinoma in lingual thyroid. The reason for this apparent predominance of follicular carcinoma is unclear. The carcinoma arising in ectopic thyroid tissue would not differ in cell types from those arising in glands occupying the normal anatomical position(8).

According to revised W.H.O. classification, papillary thyroid carcinoma (PTC) is defined as a malignant epithelial tumour showing evidence of follicular cell differentiation, typically with papillary and follicular structures as well as characteristic nuclear changes (ground glass, large size, pale irregular outline with deep grooves and pseudoinclusions)(9). Various subtypes of PTC have been identified. These include follicular, encapsulated, diffuse sclerosing, columnar cell and tall cell types. Some PTCs of follicular

variant are encapsulated (Lindsay tumour) making distinction from follicular carcinoma or adenoma difficult. However these tumours grow in a multinodular, invasive pattern with areas of sclerosis.

Chem and Rosai described the entity called ‘follicular variant of papillary carcinoma’ in 1977(9). This variant resembled papillary carcinoma in its biologic behaviour and all morphologic features with the exception that papillae were not present. The diagnosis of PTC is rendered based on the typical nuclear features. The immunophenotypic profile of follicular variant of papillary carcinoma is similar to that of classical PTC and differs from that of follicular carcinoma(10). Follicular variant also has a similar tendency for lymph node metastasis as that of classical PTC(9). In the metastatic deposit, papillary structures may be seen. The rarity of this type of tumour in lingual thyroid, along with the lack of information about its natural history and final outcome in most of the treated patients make it difficult to suggest an accurate treatment strategy or a precise prognosis(7). The same principles that dictate the decision making in the treatment of an orthotopic thyroid gland PTC applies to lingual thyroid papillary tumours including the follicular variant. All neoplasm containing foci of papillary carcinoma, regardless of proportions, have identical behaviour.

A careful review of the literature revealed 8 cases of papillary lingual thyroid carcinoma (Table 2).

Figure 6

Table 2: Table showing the features of the previously reported papillary carcinoma of the lingual thyroid

Author	Year	Age	Sex	Variant	Treatment given	Metastasis	Prognosis
Ray H(12)	1910	74	M	Not known	Surgical	Positive neck nodes	Died of another cause-3 rd year
Winslow(11)	1997	23	M	Follicular	Surgical & 131 I	Positive neck nodes	2 nd year uneventful
Jayaram G(22)	1995						
Perez JS (5)	2003	28	M	Classical	Surgical & 131 I	No metastasis	No recurrence
Goldstein B(23)	2002	48	F	Classical	Surgical & 131 I	No metastasis	Uneventful ?duration
Massine RE(4)	2001	57	M	Classical	131 I initially & surgical	Neck metastasis	Recurrence after initial 131 I treatment
Cassella A(13)	1999	66	M	Classical	Surgical & 131 I	Positive neck nodes	Not known
Singh HB(44)	1979	28	F	Classical	Surgical	None	No recurrence

One case was described as follicular variant(11). This particular case presented with a gradually enlarging neck mass arising from the thyroid gland in anatomically normal

position. Subsequent detailed assessment revealed an ectopic lingual thyroid gland. Although the thyroid mass exhibited papillary elements of papillary carcinoma, the lingual tissue was purely follicular in histologic appearance. In general, carcinomas arising from an ectopic thyroid tissue co-existing with a thyroid gland in its normal position, a metastatic origin from the primary gland is more likely. Regional neck metastasis have been documented in four of the reported cases of papillary lingual carcinoma^(4, 11,12,13). However, in our patient, there was no thyroid gland in its normal anatomical position, suggesting that the tumour is a primary lingual thyroid carcinoma.

Methods of surgical excision include lateral pharyngotomy, a transhyoid approach or a transoral approach⁽¹⁴⁾. In our patient the lateral pharyngotomy approach was used to allow for improved access to the tumour. Post operative oedema is common regardless of the approach making tracheotomy mandatory⁽⁴⁾.

High dose radionucleotide iodine 131 is recommended if positive margins exist or the disease is advanced⁽⁸⁾. Follow up scans with radioactive iodine and computerised tomography should be performed to exclude tumour recurrence.

CONCLUSION

Papillary carcinoma of the lingual thyroid is extremely rare. Radionucleotide scanning is essential to assess for the presence of thyroid tissue within the tongue and neck. Carcinoma of the lingual thyroid presents in the same manner as a symptomatic patient with a lingual thyroid. Therefore for exact pathological diagnosis a biopsy should always be taken.

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