



## A Rare Case of Squamous Cell Carcinoma in Thyroglossal Duct Cyst

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### ABSTRACT

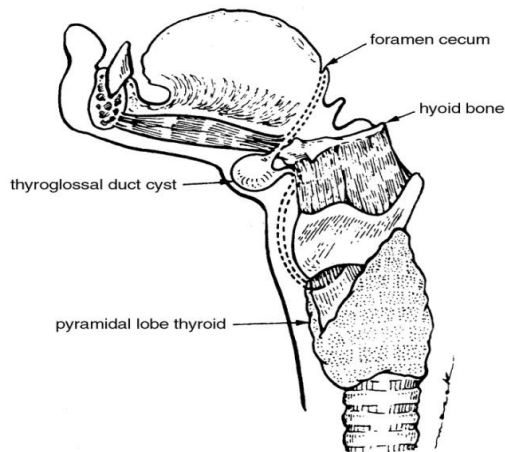
*A 35 year old female presented to our department with complaints of swelling in the neck. Radiological evaluation revealed a thyroglossal duct cyst in the anterior aspect of the neck anterior to the hyoid bone. Biopsy of the lesion was proved to be squamous cell carcinoma of squamous cell carcinoma in thyroglossal duct cyst is very rare and till date about only 14 cases were reported worldwide (1). Here we present a biopsy proven case of squamous cell carcinoma in a thyroglossal duct cyst.*

**KEY WORDS:** *Thyroglossal duct cyst, Squamous cell carcinoma, Sistrunk's operation.*

### INTRODUCTION

Thyroglossal duct cyst is a most common anomaly in thyroid development. The incidence of carcinoma arising in thyroglossal duct remnants is a well-described entity in the pathology and surgical literature, but it is less described in radiology. In this report, we discuss the radiological findings of this rare entity which aided in preoperative diagnosis, thus preventing the further progression of the disease.

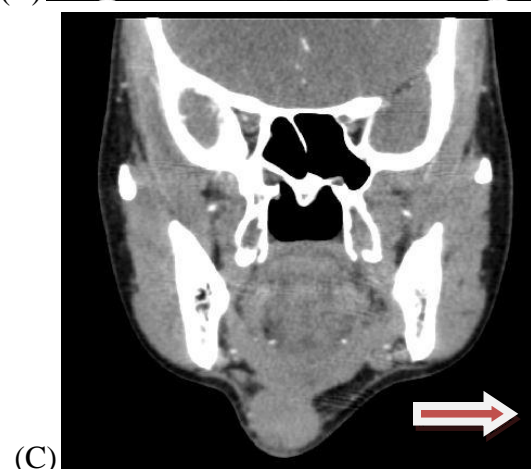
Embryologically the thyroid gland descends from the foramen caecum to a point below the thyroid cartilage. It leaves an epithelial tract known as thyroglossal tract. The tract usually fibroses then involutes during 5th-10th week of gestational life. An incomplete atrophy of this tract forms the basis of origin of the cyst. It can also be located in lingual, supra-hyoid (including sub-mental), thyrohyoid (between hyoid bone and thyroid cartilage) or suprasternal regions.



### CASE REPORT

A 35 year old female presented to our department with complaints of swelling in the neck for past 3 months, which was insidious in onset, non-progressive and associated with pain. Local examination of the swelling revealed a 2.0 x 3.0cm globular firm swelling in the midline of neck at the level of the hyoid bone, moves with deglutition of tongue. Ultrasound of the neck revealed a well defined hypoechoic lesion with internal vascularity noted in the supra thyroid region on the right side extending deeper probably into the larynx. Video laryngoscopy revealed multiple secondaries in the neck with unknown primary. FNAC of the lesion showed suspicious for malignancy. Biopsy from the neck mass was proved to be moderately differentiated squamous cell carcinoma.

CT scan revealed an irregular heterogeneously enhancing soft tissue density mass lesion of size measuring 3 x2.6x2.6 cm anterior to the hyoid bone and laryngeal cartilages. Lesion is seen to involve the strap muscles, subcutaneous fat and extending up to the skin surface (Fig A,B,C). Evidence of perilesional fat stranding extending to involve the skin. No evidence of bony erosions. Few subcentimetric sub-mental and submandibular nodes seen on the left side.



### DISCUSSION

The thyroid gland is the first endocrine gland to appear in the embryo. The thyroglossal duct is formed along the pathway of normal caudal migration of the thyroid, tethering it to the pharyngeal floor. The duct usually fibroses then involutes by 7 to 8 weeks in utero<sup>(2,3)</sup>.

Persistent remnants of the thyroglossal duct may give rise to cysts at any point along the path of descent, with 61% developing at or below the level of the hyoid bone. These cysts are lined with

columnar, cuboidal, or nonkeratinized stratified squamous epithelium. Up to 65% will also have normal thyroid tissue in the wall at sectioning<sup>(3)</sup>. It is from this tissue that most TDCa arise. Thus, 95% of TDCa are thyrogenic, with the vast majority representing papillary carcinomas. This is significant radiologically because the presence of histologic psammoma bodies and calcification in these tumors provides a feature that may be recognizable macroscopically with CT imaging.

Approximately 5% of TDCa are squamous in nature and are characteristically the most aggressive<sup>(4,5)</sup>. Virtually every type of thyroid carcinoma has been identified arising in TDC, with the exception of medullary carcinoma. This is expected because medullary carcinomas arise from parafollicular cells that develop from the ultimobranchial bodies and not the thyroid anlage<sup>(4)</sup>.

Most TDCs (70%) are diagnosed before the age of 30 years, whereas the average age of development of carcinoma is 39 years. The squamous type of TDCa tends to arise in an older age group, with an average age of incidence at 54 years<sup>(5)</sup>. Carcinoma seems to be more common in the female population, whereas benign cysts have no clear sex predilection. No predisposing factors to malignant change have been identified, although radiation is considered a risk factor<sup>(6)</sup>.

We encountered a significantly higher incidence of carcinoma arising in thyroglossal duct remnants than the reported 0.7%. Additionally, most patients with TDCs do not undergo preoperative imaging unless a suspicious clinical feature is present, such as development of a new midline mass in an adult or the rapid enlargement of a known neck mass.

The standard surgical procedure for the management of thyroglossal duct anomalies is the Sistrunk procedure, which consists of excision of the entire tract of the thyroglossal duct, the midportion of the hyoid bone, and a portion of the base of the tongue. The presence of carcinoma with cyst wall invasion or metastatic spread has been suggested by some to indicate a need for additional thyroidectomy and postoperative

irradiation<sup>(4)</sup>. There is an approximately 14% incidence of coincident thyroid gland microscopic papillary carcinoma<sup>(7)</sup>, but a reported low rate of nodal metastatic disease (7.9% compared with orthotopic thyroid papillary carcinoma with 89.8%). The best course of management, however, is still debated. If the managing surgical team advocates thyroidectomy in addition to the Sistrunk procedure, then identification of cases of carcinoma before surgery would allow for a single operation.

The imaging of TDC with sonography, CT, and, more recently, MR imaging has been well described. The most helpful feature of these cystic neck masses in the differential diagnosis is the midline location, most often at or below the hyoid bone, and the relation of the cyst to the strap muscles. The sonography literature has emphasized the variable sonographic findings with an often-complex cystic appearance<sup>(8,9)</sup>. The CT findings of a TDC have also been well documented as a well-circumscribed, low-density lesion with a thin, smooth rim. Enhancement and thickening of the cyst wall, septations, an increase in the density of the cyst contents suggest additional inflammation or infection<sup>(10-12)</sup>. A solid nodule as such in the midline of the neck may represent an ectopic or maldescended thyroid, and the lower neck should be checked for an orthotopic gland.

On MR images, a TDC may appear as a simple cyst (low T1 and high T2 signal intensity) but is most frequently of high T1 and T2 signal intensity, consistent with high protein/thyroglobulin content<sup>(13,14)</sup>. Hemorrhage within a cyst may account for variability of CT density and particularly of MR intensity, although variability was evident with both the benign TDC cases and cases of carcinoma.

There has been little in the radiology literature describing carcinoma in thyroglossal duct remnants. Several case reports have documented these rare lesions<sup>(15,16)</sup>, and the literature has suggested that the presence of solid components in a TDC should raise the suspicion of malignancy<sup>(17)</sup>. Inflammatory processes, however,

may show thickening of the cyst wall and the presence of solid components<sup>(10-12)</sup>.

In our case ultra sound and the CT of the neck revealed a large nodular lesion in the anterior aspect of the neck in the subcutaneous plane. The lesion was situated anterior to the hyoid bone and thyroid cartilage in the midline. There was evidence of perilesional fat stranding extending to involve the skin. Further post-op tissue biopsy revealed squamous cell carcinoma of the thyroglossal duct cyst.

To our knowledge, calcification has not been reported in association with TDCs, even in the presence of chronic inflammation. Calcification seems to be a specific, although not sensitive, indicator of papillary carcinoma, the most common type of TDCa.

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