

# PRIMARY PAPILLARY CARCINOMA IN THYROGLOSSAL DUCT CYST

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**Abstract-** Thyroglossal duct cysts are the most common congenital anomalies in thyroid development, which in less than 1% of cases are malignant. In most cases the diagnosis is made postoperatively. Up to now, a few cases have been reported which had been papillary carcinoma. Controversies exist concerning its nature and treatment. We present a 30 years old woman with papillary carcinoma of thyroglossal duct cyst, identified in pathologic study after sistrunk procedure. In our case there was neither invasion to adjacent tissue nor lymph node involvement. No further procedure was performed. After 2 years of followup, the patient was asymptomatic and there was no evidence of recurrence. Based on otolaryngologic references, we recommend sistrunk procedure and long term follow up for management of thyroglossal duct cyst with papillary carcinomas without marginal invasion and node metastasis.

*Acta Medica Iranica* 2007; 45(3): 236-238.

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**Key words:** Thyroglossal cyst, Papillary carcinoma, Surgery, Sistrunk procedure

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## INTRODUCTION

Thyroglossal duct cysts are the most common congenital cervical abnormalities in childhood and thyroglossal duct cysts (TDC) are the most common midline neck mass and the most common congenital anomalies in thyroid development (1, 2). The thyroid gland develops from the floor of the primitive pharynx during the third week (3). The thyroid gland descends from its origin in 4th week of embryogenesis (3). During this descent, the connection between thyroid gland and foramen cecum may persist to form thyroglossal duct (1). Thyroglossal duct remnant may be present in approximately 1% of cases (2, 4). They are usually benign, but approximately 1% of them may develop neoplastic changes (1). Carcinoma arising in TDC is

rare and characterised by a relatively non aggressive behaviour with rare lymphatic spread.

The most common histopathological type is papillary adenocarcinoma, accounting for roughly 85% to 92% of these tumors (1). Regional lymph node metastasis of papillary carcinomas in thyroglossal cysts occurs in 7.7-12.9% of cases. Up to now 150 cases of TDC carcinoma, predominantly papillary type, have been reported (5). Squamous cell carcinoma is the second most frequently reported cancer and accounts for 7% of cases, being more aggressive with worse prognosis. Mixed papillary and squamous cell carcinoma are described as extremely rare conditions. Medullary carcinoma has never been found in a thyroglossal duct carcinoma (2) and hurtle cell has been reported only in one case. Most cases of thyroglossal duct cyst carcinoma have been diagnosed in 3<sup>rd</sup>-4<sup>th</sup> decade of life and it has been reported rarely in children before 14 years old (2). Here we report a case of a 30 years old woman with papillary carcinoma of thyroglossal duct cyst.

Received: 23 Jun. 2004, Revised: 24 Apr. 2005, Accepted: 24 May 2005

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## CASE REPORT

The patient was a 30 years old woman with anterior midline neck mass. She had no other evidence of disease.

On physical examination there was a 3.5 cm diameter smooth, painless, cystic nodule beneath the hyoid bone. The thyroid gland was normal on palpation. There was neither cervical lymphadenopathy nor other neck mass. Direct laryngoscopy, fine needle aspiration biopsy and cervical computed tomography (CT) scan were normal and compatible with the physical findings at a thyroglossal duct cyst (Fig. 1). Thyroid scan revealed no functional tissue in the mass.

Laboratory investigations revealed normal T3, T4 and TSH levels. The patient underwent surgery with this diagnosis and the mass was resected by the usual Sistrunk procedure. There was no local sign of invasion to tissues surrounding the cyst or duct at surgery.

The patient was discharged within 24 hours. Histologic section showed a papillary carcinoma in the thyroid tissue of the thyroglossal cyst with normal thyroid tissue at the boundary of carcinoma (Fig. 2). There was no capsular invasion and margins were normal.

The patient was followed up for two years with no further evidence of disease recurrence. Informed consent was obtained from the patient for publication of his details in this report.



**Fig. 1.** Cervical computed tomography (CT) scan of the patient which was normal and compatible thyroglossal duct cyst.



**Fig. 2.** Histologic section showing a papillary carcinoma in the thyroid tissue of the thyroglossal cyst with normal thyroid tissue at the boundary of carcinoma.

## DISCUSSION

TDC carcinoma is very rare and only a few cases have been reported. Risk factors of thyroid carcinoma include history of ionizing radiation, history of thyroid diseases, age, tumor size, tumor spread and histopathological factors (6).

Criteria for diagnosis of primary carcinoma of the thyroglossal duct are: histological identification of a thyroglossal duct cyst by demonstrating the duct or cyst with an epithelial consisting of normal thyroid follicles within the wall of the cysts. Normal thyroid tissue is adjacent to the tumor and normal thyroid gland without primary carcinoma in histopathological examination (1).

Kennedy *et al.* found three cases of papillary carcinoma and one case of squamous cell carcinoma in a large review of 115 thyroglossal duct cysts (7). Neoplasms might have arisen from these thyroid remnants or the lining epithelium. With the possible exception of medullary carcinoma, all other thyroid malignancies have been documented to occur in thyroglossal duct cyst (8). Papillary carcinoma (75–85%), mixed papillary-follicular carcinoma (7%) and squamous cell carcinoma (5%) constitute the majority of malignant lesions developing in thyroglossal duct cyst. Other histologic subtypes such as follicular carcinoma and Hurtle cell carcinoma have been reported rarely in the literature. Squamous cell carcinoma has the least favorable prognosis (8). In our case, the presence of carcinoma within a thyroglossal duct, normal thyroid tissue within the mass and normal scintigraphic

examination of the thyroid gland support the diagnosis of papillary carcinoma arising in a thyroglossal duct remnant rather than a metastasis from a primary carcinoma of the thyroid gland.

Diagnosis of carcinoma of the thyroglossal duct is usually made on histological examination. The histological diagnosis of thyroglossal duct carcinoma requires the presence of malignant cells and also the presence of normal others. The reported incidence of TDC carcinoma is approximately 1% (1, 2, 4). Imaging techniques (ultrasonogram, scintigraphy and CT) are usually unable to diagnose malignant disease preoperatively (1). Fine needle aspiration yields a correct result in only 66% of cases. Surgical treatment of these carcinomas is controversial. A small number of cases have been reported; according to Patel (9) univariate analysis of 62 cases the Sistrunk procedure is adequate for most patients with incidentally diagnosed TDC carcinoma in the presence of clinically and radiologically normal thyroid gland. Another review concluded Sistrunk operation and low rates of complication (9.08%) and recurrence (1.82%) (10). Papillary carcinoma is the most common histologic subtype, with a slight female preponderance (female:male = 3:2) (11). Papillary carcinoma within thyroglossal duct cyst usually presents as an asymptomatic mass. Metastatic lymphadenopathy at presentation has been reported in 1% of patients. There is still controversy about thyroid removal for a papillary carcinoma, but all patients should receive suppressive doses of thyroid hormone.

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