

Primary papillary carcinoma in a thyroglossal duct cyst

Abstract

Thyroglossal duct cysts (TDC) are the most common congenital anomalies of the thyroid gland and less than 1% of them become malignant. The malignancy most often found is papillary carcinoma. The diagnosis is usually made postoperatively. Controversy exists whether TDC are primary or secondary and also as for their treatment regime. In this case report, we present a 30-year-old woman with papillary carcinoma of the TDC, identified only by pathology after surgical ablation by Sistrunk's procedure. Invasion of the carcinoma to the adjacent tissues or lymph node metastases were not found. Thyroid function tests were normal as well as the thyroid scan. After a two-year follow up, the patient remained asymptomatic with no evidence of recurrence, as tested clinically and by ultrasonography. Since our patient had no marginal invasion of the carcinoma in the surrounding tissues or lymph node metastases, we recommended the Sistrunk's surgical procedure, suppressive treatment by thyroid hormones and long term follow-up.

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Introduction

Thyroglossal duct cyst (TDC) is the most common congenital anomaly of the thyroid gland, the most common midline neck mass [1,2] and also the most common congenital cervical abnormality in childhood. Seventy percent of all congenital cervical abnormalities in childhood and 7% in adults are due to TDC [1-3]. Carcinomas arising from a TDC are rare, only 1% of the TDC cases, and characterised by a relatively non aggressive behaviour and by rare lymphatic spread [2]. About 85%-92% of all TDC carcinomas are papillary carcinomas [2]. Regional lymph node metastases of these carcinomas of the TDC occur in 7.7%-12.9% of the cases [2]. To our knowledge, 226 cases of TDC have been reported in the English literature up to now.

We present this case of TDC with papillary carcinoma because of the rarity of such cases, the negative pre-surgery diagnosis, the interesting clinical and laboratory findings and the satisfactory after two years results of the mild treatment regime that we applied.

Case presentation

The patient was a 30-year-old woman with an 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 another neck mass. Direct laryngoscopy and fine needle aspiration biopsy of the mass showed no evidence of malignancy. The computerized tomography (CT) scan of the cervix was also negative (Fig. 1). Thyroid scan with technetium pertechnetate revealed no functional tissue in the neck mass (Fig. 2). Serum thyroxine (T_4), triiodothyronine (T_3) and thyroid stimulating hormone (TSH) were within normal range. A TDC was diagnosed, the patient underwent surgery and the mass was resected by Sistrunk's surgical procedure. This procedure requires excision not only of the cyst but also of the duct and its branches. The intimate association of the duct with the hyoid bone mandates simultaneous removal of the central portion of the hyoid bone to ensure complete removal of the duct. Histology showed a papillary carcinoma in the thyroid tissue of the TDC with normal thyroid tissue at the boundaries of the carcinoma. There was no capsular invasion in the thyroid gland. There was no local sign of cancer invasion to the surrounding tissues or to the duct of the TDC. The patient was followed up for two years with no further evidence of disease recurrence. The follow up of the patient consisted of clinical examination and ultrasonography (US) of the surgical region and the thyroid gland. In addition, suppressive treatment was suggested.

Discussion

The thyroid gland descends from the floor of the primitive pharynx during the fourth week of embryogenesis [4,5]. During this descent the connection between the thyroid gland and the foramen cecum may persist and form the thyroglossal duct [2]. Most cases of TDC carcinoma

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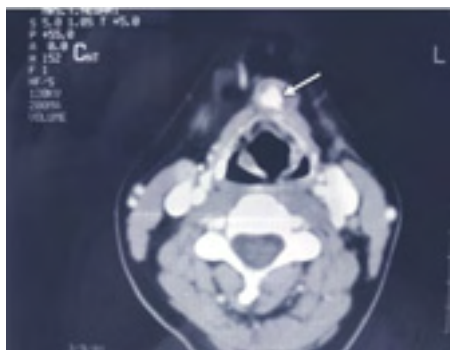


Figure 1. Contrast enhanced CT scan. There is normal thyroid tissue mass in the thyroglossal duct (arrow)

have been diagnosed during the third and fourth decades of life and rarely in children before 14 years of age [4,5]. TDC carcinoma is rare. A rapid increase in the size, pain and presence of enlarged lymph nodes may suggest malignancy of TDC [4,5]. Kennedy et al. (1998) in a large review of 115 TDC cases, found three cases of papillary carcinoma and one case of squamous cell carcinoma [6]. These neoplasms may arise either from thyroid remnants or from the lining epithelium of the TDC. With the possible exception of medullary carcinoma, all other thyroid malignancies have been described to occur in TDC [7]. Papillary carcinoma (75%–85%), mixed papillary-follicular carcinoma (7%) and squamous cell carcinoma (5%) constitute the majority of malignant lesions developing in TDC [7]. Other histologic subtypes such as follicular carcinoma and Hürtle cell carcinoma have been rarely reported in the literature. From all the above, squamous cell carcinoma has the least favorable prognosis [8].

Imaging techniques (US, scintigraphy and CT) are usually unable to diagnose malignant disease preoperatively [2]. Fine needle aspiration of the mass yields a correct result in only 66% of the cases [9].

In our case, the presence of papillary carcinoma within the thyroglossal duct, with normal thyroid tissue around the carcinoma, normal thyroid scintigraphy, and CT scan support the view of a papillary carcinoma in situ.

Surgical treatment of these carcinomas is controversial. A small number of cases have been reported. According to Patel et al. (2002) univariate analysis of 62 cases showed that the Sistrunk's surgical procedure is adequate for most patients with incidentally diagnosed TDC carcinoma in the presence of clinically and radiologically normal thyroid gland [8]. Another review concluded that Sistrunk's operation induced a low rate of complications and recurrences, 9.08% and 1.82% respectively [10].

Malignant foci in the thyroid could be either primary or metastatic from a primary carcinoma in the TDC. There is still controversy about the removal of the thyroid gland in case of papillary carcinoma of the TDC [11]. If the thyroid gland is nodular with a cold nodule present on the thyroid scan, if there are enlarged neck lymph nodes or if there is a history of neck irradiation, thyroidectomy is recommended [12]. In our case there was nothing of the above and so the thyroid gland was spared.

There is a slight female to male preponderance, 3:2 [13].

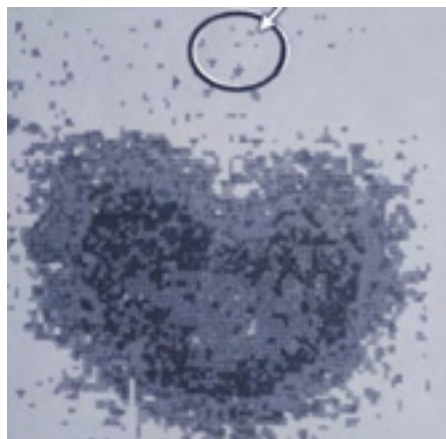


Figure 2. The thyroid scan shows relatively homogeneous radiotracer uptake throughout the gland. Thyroid gland is also normal in size, shape and in location. The palpable mass in the anterior aspect of the neck reveals no tracer activity (arrow)

Papillary carcinoma within TDC is usually presented as an asymptomatic mass with metastatic lymphadenopathy in 1% of the patients. Suppressive doses of thyroid hormone are recommended for a long time after the operation [11].

In conclusion, this case has been presented because it is rare, the carcinoma was not diagnosed preoperatively, the thyroid gland was not removed and the patient was free of any signs of carcinoma after two years.

Bibliography

1. Fernandez JF, Ordoñez NG, Schultz PN, et al. Thyroglossal duct carcinoma. *Surgery* 1991; 110: 928-935.
2. Martin-Perez E, Larranaga E, Marron C. Primary papillary carcinoma arising in a thyroglossal duct cyst. *Eur J Surg* 1997; 163: 143-145.
3. Dedivitis RA, Guimaraes AV. Papillary thyroid carcinoma in thyroglossal duct cyst. *Int Surg* 2000; 85: 198-201.
4. Bhagavan BS, Rao DRG, Weinberg T. Carcinoma of the thyroglossal duct cyst: Case reports and review of the literature. *Surgery* 1970; 67: 281-292.
5. Jameson JL, Weetman AP. Disorders of the thyroid gland. In: Braunwald E, Fauci AS, Kasper DL, et al. *Harrison's Principles of Internal Medicine*. 15th edn. New York: Mc Graw Hill; 2001: p. 2060.
6. Kennedy TL, Whitaker M, Wadhi G. Thyroglossal duct carcinoma: a rational approach to management. *Laryngoscope* 1998; 108(8 pt 1): 1154-1158.
7. Ozturk O, Demirci L, Egeli E, et al. Papillary carcinoma of the thyroglossal duct cyst in childhood. *Eur Arch Otorhinolaryngol* 2003; 260: 541-543.
8. Patel SG, Escrig M, Shaha AR, et al. Management of well-differentiated thyroid carcinoma presenting within a thyroglossal duct cyst. *J Surg Oncol* 2002; 7: 134-139.
9. Bardales RH, Suhrlund MJ, Korourian S, et al. Cytologic findings in thyroglossal duct carcinoma. *Am J Clin Pathol* 1996; 106:615-619.
10. Dedivitis RA, Camargo DL, Peixoto GL, et al. Thyroglossal duct: a review of 55 cases. *J Am Coll Surg* 2002; 194: 274-277.
11. Hilgar AW, Thomson SD, Smallman LA, et al. Papillary carcinoma arising in a thyroglossal duct cyst: a case report and literature review. *J Laryngol Otol* 1995; 109: 1124-1127.
12. Sebastian BJ, Sreekumar S, Vaidyanathan S. Thyroglossal duct cyst carcinoma- report of a case and review of literature. *Thyroid Research and Practice* 2004; 1: 27-29.
13. Weiss SD, Orlich CC. Primary papillary carcinoma of a thyroglossal duct cyst. *Br J Surg* 1991; 78: 87-89.

