Case Report-2

Primary Papillary Carcinoma Arising in a Thyroglossal Duct Cyst

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ABSTRACT

Well-differentiated thyroid carcinoma (WDTC) is diagnosed in approximately 1.5% of thyroglossal duct cysts. There is no clear consensus regarding further management after adequate excision of the cyst. We report a case of primary papillary carcinoma arising in the thyroglossal duct cyst. Patient underwent excision of thyroglossal cyst followed by total thyroidectomy.

Conclusion: The strategies adopted need to be individualized that may require adequate surgery of thyroglossal duct cyst (TGDC) only or aggressive treatment in form of additional thyroid surgery, radiiodine therapy and suppressive dose of thyroxin.

INTRODUCTION

Thyroglossal duct cyst is one of the commonest congenital midline abnormality. Carcinoma within cyst is rare. Brentano gave its first description in 1911 and since than about 200 cases have been reported in the literature. On histopathology- Papillary carcinoma predominates. The prognosis of papillary carcinoma arising in a thyroglossal cyst is similar to that of papillary carcinoma of thyroid gland having cure rates in excess of 95%. There is lack of consensus in literature regarding optimal management of this condition. It is due to lack of pre-operative diagnosis, rarity of the condition and possibility of associated thyroid malignancy. We present one such case.

Case: A 31-year-old woman presented with painless swelling located anterior to midline of neck of 2 months duration. There were no associated symptoms of hoarseness of voice, fever and discharge from swelling. There were no features suggestive of hyper or hypothyroidism. On examination – swelling was visible in upper part of the neck and was moving on deglutition and protrusion of tongue. Overlying skin was normal with no visible dilated veins. It was firm and cystic in nature, non-tender and smooth, with a size 2 X 2 cm below hyoid bone.

Indirect laryngoscopy examination was unremarkable. Routine hematological, biochemical tests and chest x-ray were normal. Thyroid scintigram done with Tc 99m-pertechnetate was normal. Ultrasound scan of neck revealed small hypo-echoic nodule measuring 10x 9 mm in size in suprahypoid area with the possibility of thyroglossal duct cyst while both the lobes and isthmus of thyroid gland were normal. Fine needle aspiration...
cytology revealed papillary carcinoma. Excision of thyroglossal cyst was carried out and there was no evidence of lymph node enlargement. Histopathology of specimen revealed papillary carcinoma with no soft tissue invasion. (Fig. 1)

Patient underwent total thyroidectomy to eliminate any malignant focus in the thyroid. Histological examination of thyroid gland revealed normal colloid filled material with no evidence of malignancy. There was no regional lymphadenopathy. These findings suggested that tumour was a primary papillary carcinoma arising in a thyroglossal cyst. The patient was put on suppressive dose of thyroxin.

The clinical examination and radiological investigations for the thyroid gland in this patient were normal and though the risk of cancer in thyroid gland was very small, total thyroidectomy was carried out in this case keeping in mind the poor follow-up by the patient.

**DISCUSSION**

Thyroglossal duct remnants have been found in up to 7% of adults.\(^4\) There has been some speculation in the literature regarding the source of well differentiated thyroid carcinoma (WDTC) in TGDC, but thyroid tissue has been histological demonstrated in up to two-thirds of TGDC,\(^5\) and malignant transformation is possible just as in the thyroid gland. The incidence of WDTC in TGDC is approximately 1.6% as reported in two large series.\(^5,6\) Though the management of TGDC itself is relatively straightforward, whether additional treatment is necessary to treat further is controversial.

Preoperative evaluation of thyroglossal duct cyst includes a thorough head and neck examination, palpation of thyroid gland and thyroid function tests. Thyroid scan should be done if there is a suspicion of an ectopic thyroid gland. It is also indicated if a mass is palpable within the cyst, thyroid gland or in the neck.\(^2\) An ultrasound examination may aid in the diagnosis of malignancy by demonstrating a mural nodule, calcification or lymph node metastases.\(^7\) The diagnosis is usually based on pathological examination of the cyst.\(^8\) Papillary carcinoma is the most common histological subtype. It is difficult to assess the incidence of carcinoma in the thyroid gland concomitant with TGDC carcinoma as not all patients undergo total thyroidectomy along with excision of TGDC. Snehel et al found an incidence of 25% WDTC in thyroid specimen in a review.\(^9\) The impact of finding microscopic foci of WDTC in an otherwise normal thyroid gland has been debated extensively. It has been demonstrated that there is no significantly altered outcome in such patients. There is no data to support the routine elective nodal dissection, but suspicious nodes encountered at surgery must be dissected and analyzed.

Recurrence rates are lowest when total ablation of thyroid tissue is achieved therefore thyroidectomy followed by radioactive iodine ablation should be considered especially if tumour recurrence or metastases occurs.\(^2\) Thyroid suppression is recommended for all patients with papillary carcinoma of thyroglossal duct cyst regardless of the presence of a normal thyroid scan or whether the patient had thyroidectomy or not.\(^2,10\) Literature showed that a total thyroidectomy has been performed consequent to the diagnosis of the thyroglossal duct cyst carcinoma in most of the patients.
This case is reported in view of its rarity and to highlight the therapeutic options in the management of malignancy arising in thyroglossal duct cysts.

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