

Case report

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Inapparent twin malignancy in thyroglossal cyst: Case Report

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Published: 27 August 2003

Received: 02 July 2003

World Journal of Surgical Oncology 2003, 1:15

Accepted: 27 August 2003

This article is available from: <http://www.wjso.com/content/1/1/15>

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Abstract

Background: Although malignancy has been reported in thyroglossal cysts, synchronous occurrence of two malignancies is extremely rare.

Case Report: A case of concurrent papillary and squamous carcinoma arising in the thyroglossal cyst is presented here.

Conclusions: Papillary and squamous carcinoma simultaneously occurring in a thyroglossal cyst is rarely diagnosed prior to surgery and pose a therapeutic dilemma. In view of the extreme rarity of the condition, controversies do exist regarding the optimal strategy to be adopted. The ideal procedure needs to be individually tailored and involve a combination of surgery, radio ablation, thyroid suppression and external radiotherapy.

Introduction

Malignant neoplasms rarely arise in thyroglossal cysts [1,2]. Papillary carcinoma predominates among them. Squamous carcinoma accounts for only 5 % of the cases [3]. Only two cases of concurrent papillary and squamous carcinoma have been reported so far [4,5]. Lack of an accurate preoperative diagnosis, rarity of the condition, and the possibility of an associated thyroid malignancy duly contribute to the confusion regarding the optimal management of this condition. We present one such case and discuss the various therapeutic options.

Case Report

A 48-year-old man presented with a swelling in the front of neck for past one year associated with a recent increase

in size and hoarseness of voice. On examination, he was found to have a 5 × 6 cm cystic, non-transilluminant swelling below the hyoid, which moved with deglutition and protrusion of tongue but had restricted intrinsic mobility. Indirect laryngoscopic examination was unremarkable. Fine needle aspiration cytology was carried out which revealed thyroglossal cyst. Routine hematological and biochemical investigations and chest X roentgenogram were normal.

With a preoperative diagnosis of thyroglossal cyst, Sistrunk's procedure was planned. On exploration of the neck at surgery he was found to have two cystic swellings intimately related to each other, one was in the midline and the other was laterally placed. The latter was

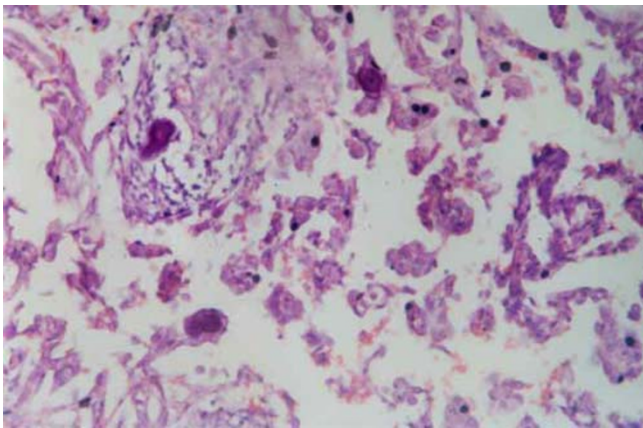


Figure 1
photomicrograph showing tumor cells with uniform opened out nucleus arranged in a papillary pattern. Psammoma bodies are seen. [Hematoxylin and Eosin $\times 200$]

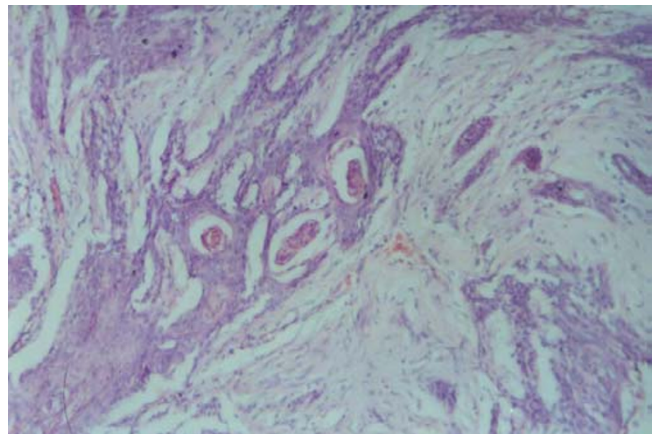


Figure 2
photomicrograph showing sheets of tumor cells with eosinophilic cytoplasm and hyperchromatic nucleus. Keratin pearl formation is seen. [Hematoxylin and Eosin $\times 200$]

intimately related to the thyroid gland and adherent to the carotid sheath, strap muscles and the laryngeal apparatus. There was no regional lymphadenopathy. The swelling was meticulously dissected free from the carotid sheath and the laryngeal apparatus and was excised along with a portion of the strap muscles. A near total thyroidectomy was also performed. The right recurrent laryngeal nerve could not be identified at surgery and was presumed sacrificed during the procedure. The patient developed stridor in the immediate postoperative period, which necessitated endotracheal re-intubation but resolved later with conservative measures.

Histopathology of the resected specimen showed distinct foci of papillary carcinoma ([Figure 1) and squamous carcinoma (Figure 2) in the lateral cystic swelling and a normal thyroid gland. The midline swelling was found to have inflammatory tissue only. The patient was put on suppressive dose of thyroxin. Thyroid scintigraphy, radioiodine ablation of remnant thyroid and external beam radiotherapy to the tumor bed was planned however; the patient was lost to follow-up.

Discussion

Thyroglossal cysts are the most common non-odontogenic cyst in the neck and are presents in approximately 7% of the general population [1,6]. Primary carcinoma, on the other hand, is rare and is seen in less than 1% of cases [1,2]. Although rapid increase in size, dysphagia, hoarseness of voice and pain may be harbingers of malignancy, the diagnosis is usually based on pathological examination of the cyst [1,7-12]. Preoperative evaluation of thyroglossal duct cyst includes a thorough head and

neck examination, palpation of thyroid gland, thyroid function tests and a thyroid scan if there is a suspicion of an ectopic thyroid gland or a mass is palpable within the cyst, thyroid gland or in the neck [9]. An ultrasound examination may aid in the diagnosis of malignancy by demonstrating a mural nodule, calcification or lymph node metastases [13]. Computerized tomography has also been used for the same purpose but a previously infected cyst with thickened walls, presence of ectopic thyroid tissue within the cyst as well as a congenital dermoid cyst with debris may mimic malignancy [14]. Papillary carcinoma accounts for more than 90% of all cases and squamous carcinoma is seen in only 5% of cases [3]. Concurrent papillary and squamous carcinoma arising in thyroglossal cyst is extremely rare and only two cases have been reported so far [4,5]. In the case described, as the foci of papillary and squamous carcinoma are separate and distinct, it is unlikely that the latter arose from squamous metaplasia of the papillary carcinomatous component.

The prognosis of papillary carcinoma arising in a thyroglossal cyst is good and is similar to that of papillary carcinoma of thyroid gland having cure rates in excess of 95% [9,15]. Sistrunk's procedure is preferred to simple excision and would suffice if the thyroid gland was found to be normal and negative margins can be achieved [16]. If the thyroid and lymph nodes are grossly normal during the procedure, a postoperative thyroid scan is recommended. However, if thyroid nodules are palpable preoperatively, or the pre or postoperative thyroid scan is abnormal, thyroidectomy is warranted [17,18]. In the present case, as the lesion was intimately related to the thyroid gland and given the 10 - 14% risk of another

focus of carcinoma in the thyroid gland, we decided to carry out a near total thyroidectomy. Lymphadenopathy on the other hand is found only in 8% of the cases, hence only the significant involvement of regional lymph nodes necessitates a modified radical neck dissection [3,10]. Central compartment dissection should also be considered in view of the fact that these lymph nodes may be involved in the absence of concomitant foci of malignancy in the thyroid gland [13]. Recurrence rates are lowest when total ablation of thyroid tissue is achieved and therefore thyroidectomy followed by radioiodine ablation should be considered especially if tumor recurrence or metastasis occurs [9]. Ultrasound examination of the neck as well as plasma thyroglobulin estimation may be helpful in detecting recurrence. Thyroid suppression is recommended for all patients with papillary carcinoma of thyroglossal cyst regardless of the presence of a normal thyroid scan or whether the patient had a thyroidectomy or not [9,11,19,20].

Patients with squamous carcinoma arising in a thyroglossal cyst have a poorer prognosis and therefore should be treated with Sistrunk's procedure and postoperative external beam radiotherapy to both sides of the neck even in the absence of regional lymphadenopathy [9,21].

This case is reported in view of the extreme rarity and to highlight the therapeutic options in the management of malignancies arising in thyroglossal cysts.

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