

Case report

Open Access

Inapparent twin malignancy in thyroglossal cyst: Case Report

Ramachandran Vijay*¹, Karattuthazhath Karimbayil Rajan¹ and Moosa Feroze²

Address: ¹Department of General Surgery, Calicut Medical College, Kozhikkode, Kerala, India and ²Department of Pathology, Calicut Medical College, Kozhikkode, Kerala, India

Email: Ramachandran Vijay* - divijayr1@yahoo.com; Karattuthazhath Karimbayil Rajan - drkkrcrlt@sancharnet.in; Moosa Feroze - mferoze@sify.com

* Corresponding author

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>

© 2003 Ramachandran et al; licensee BioMed Central Ltd. This is an Open Access article: verbatim copying and redistribution of this article are permitted in all media for any purpose, provided this notice is preserved along with the article's original URL.

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

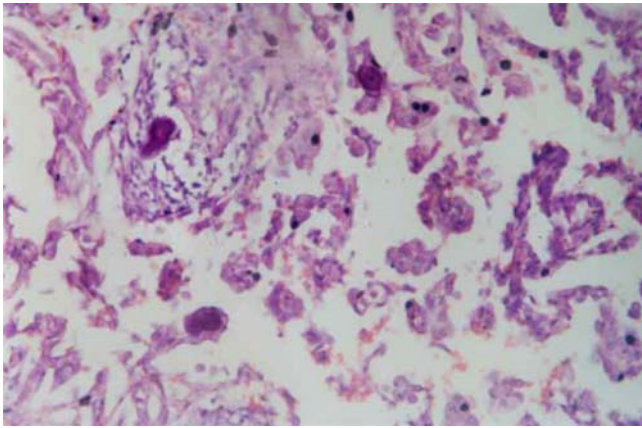


Figure 1
photomicrograph showing tumor cells with uniform opened out nucleus arranged in a papillary pattern. Psammoma bodies are seen. [Hematoxylin and Eosin $\times 200$]

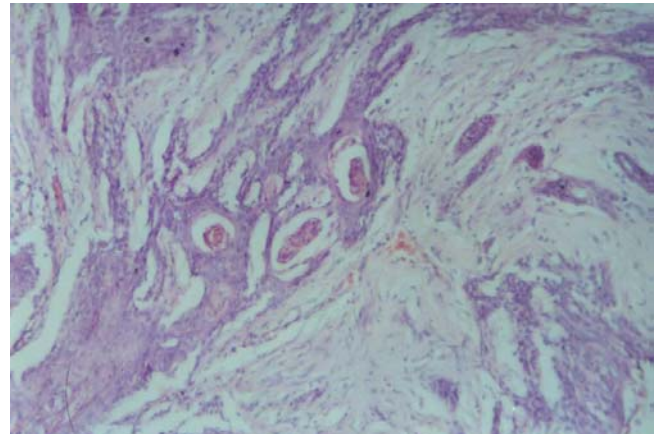


Figure 2
photomicrograph showing sheets of tumor cells with eosinophilic cytoplasm and hyperchromatic nucleus. Keratin pearl formation is seen. [Hematoxylin and Eosin $\times 200$]

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 present 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.

References

- Allard RHB: **The thyroglossal cyst.** *Head Neck Surg* 1982, **5**:134-146.
- Nussbaum M, Buchwald RP, Ribner A, Mori K and Litwins J: **Anaplastic carcinoma arising from median ectopic thyroid (thyroglossal duct remnant).** *Cancer* 1981, **48**:2724-2728.
- Renard TH, Choucair RJ, Stevenson WD, Brooks WC and Poulos E: **Carcinoma of the thyroglossal duct.** *Surg Gynecol Obstet* 1990, **171**:305-308.
- Ronan SG, Deutsch E and Ghosh L: **Thyroglossal duct carcinoma: light and electron microscopic studies.** *Head Neck Surg* 1986, **8**:222-225.
- Kwan WB, Liu FF, Banerjee D, Rotstein LE and Tsang RW: **Concurrent papillary and squamous carcinoma in a thyroglossal duct cyst: A case report.** *Can J Surg* 1996, **39**:328-332.
- Ellis P and van Nostrand AW: **The applied anatomy of thyroglossal tract remnants.** *Laryngoscope* 1977, **87**:765-770.
- Bhagavan VS, Rao DR and Weinberg T: **Carcinoma of thyroglossal duct cyst: Case report and review of the literature.** *Surgery* 1970, **67**:281-292.
- Yildiz K, Koksall H, Ozpran Y, Muhtar H and Telatar M: **Papillary carcinoma in a thyroglossal duct remnant with normal thyroid gland.** *J Laryngol Otol* 1993, **107**:1174-1176.
- LaRouere MJ, Drake AF, Baker SR, Richter HJ and Magielski JE: **Evaluation and management of a carcinoma arising in a thyroglossal duct cyst.** *Am J Otolaryngol* 1987, **8**:351-355.
- Martin Perez M, Burguno Garcia M, Echevarria Iturbe C and Martinez Perez D: **Papillary carcinoma arising in a thyroglossal duct cyst: Report of two cases.** *J Oral Maxillofac Surg* 1993, **51**:89-93.
- Hilger AVW, Thompson SD, Smallman LA and Watkinson JC: **Papillary carcinoma arising in a thyroglossal duct cyst: A case report and literature review.** *J Laryngol Otol* 1995, **109**:1124-1127.
- Jacques DA, Chambers RG and Oertel JE: **Thyroglossal tract carcinoma: A review of the literature and addition of eighteen cases.** *Am J Surg* 1970, **120**:439-446.
- Cignarelli M, Ambrosi A, Marino A, Lamacchia O, Cincione R and Neri V: **Three cases of papillary carcinoma and three of adenoma in thyroglossal duct cysts: clinical – diagnostic comparison with benign thyroglossal duct cysts.** *J Endocrinol Invest* 2002, **25**:947-954.
- Branstetter BF, Weismann JL, Kennedy TL and Whitaker M: **The CT appearance of thyroglossal duct carcinoma.** *Am J Neuroradiol* 2000, **21**:1547-1550.
- Schlumberger MJ: **Papillary and follicular thyroid carcinoma.** *N Engl J Med* 1998, **338**:297-306.
- McNicol MP, Hawkins DB, England K, Penny R and Maceri DR: **Papillary carcinoma arising in a thyroglossal duct cyst.** *Otolaryngol Head Neck Surg* 1989, **99**:50-54.
- Chen F, Sheridan B and Nankervis J: **Carcinoma of the thyroglossal duct: Case reports and a literature review.** *Aust NZ J Surg* 1993, **63**:614-616.
- Weiss SD and Orlich CC: **Primary papillary carcinoma of a thyroglossal duct cyst: Report of a case and a literature review.** *Br J Surg* 1991, **78**:87-89.
- Kum CK, Goh P and Teh M: **Papillary carcinoma arising in a thyroglossal duct.** *Aust NZ J Surg* 1993, **63**:738-740.
- Maziak D, Borowy ZJ, Deitel M, Jaksic T and Ralph-Edwards A: **Management of papillary carcinoma arising in thyroglossal duct anlage.** *Can J Surg* 1992, **35**:522-525.
- Bosch K, Kummer E and Hohmann F: **Carcinoma of the thyroglossal duct.** *Neth J Surg* 1986, **38**:36-40.

Publish with **BioMed Central** and every scientist can read your work free of charge

"BioMed Central will be the most significant development for disseminating the results of biomedical research in our lifetime."

Sir Paul Nurse, Cancer Research UK

Your research papers will be:

- available free of charge to the entire biomedical community
- peer reviewed and published immediately upon acceptance
- cited in PubMed and archived on PubMed Central
- yours — you keep the copyright

Submit your manuscript here:
http://www.biomedcentral.com/info/publishing_adv.asp

