

Simultaneous typical carcinoid tumour of larynx and occult papillary thyroid carcinoma

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Abstract

Typical carcinoid tumour of the larynx is extremely rare. We report the case of a 43-year-old man initially diagnosed with typical carcinoid tumour of the larynx, presenting with a foreign body sensation in his throat. Coincidentally, an occult papillary thyroid carcinoma was diagnosed, following detection of a suspicious lymph node on pre-operative imaging studies, and confirmation by neck dissection and subsequent total thyroidectomy. Following surgery and radioiodine therapy for these cancers, the patient was alive, well and free of disease two years later. Typical carcinoid tumour of the larynx with a synchronous occult papillary thyroid carcinoma has not previously been reported. The optimal treatment strategy for this case is also discussed.

Key words: Synchronous Neoplasms; Larynx; Papillary Carcinoma; Carcinoid Tumour; Thyroid Neoplasms

Introduction

Neuroendocrine carcinoma of the larynx is very rare. It can be divided into: typical carcinoid tumour (i.e. well differentiated neuroendocrine carcinoma); atypical carcinoid tumour (i.e. moderately differentiated neuroendocrine carcinoma); and small cell carcinoma (i.e. small neuroendocrine carcinoma).¹ Small cell carcinoma has the poorest prognosis, even after multimodal treatments have been instituted. Atypical carcinoid tumour, which is the most common type, can be cured with radical surgery. Typical carcinoid tumour, which is extremely rare, is less aggressive and generally localised, with rare regional or distant metastasis. Local excision is adequate for the treatment of a typical carcinoid tumour of the larynx.²

It has been reported that neuroendocrine carcinoma can metastasise to the thyroid gland, mimicking a medullary thyroid carcinoma.³ However, simultaneous papillary thyroid carcinoma has not been previously reported. The following secondary primary cancers are known to coexist with papillary thyroid carcinoma: squamous cell carcinoma, Ewing's sarcoma and osteosarcoma.^{4–6}

In this paper, we report a case of typical carcinoid tumour of the larynx, with simultaneous papillary thyroid carcinoma. The latter tumour was incidentally detected from a metastatic lymph node revealed by imaging studies and was confirmed by neck dissection and subsequent thyroidectomy. To our knowledge, this is the first published description of such a case.

Case report

A 43-year-old man, who had smoked tobacco for 20 years, presented with an eight-month history of a foreign body

sensation in the throat, unrelated to swallowing or speaking.

Laryngoscopy revealed a reddish lesion on the superior surface of the right arytenoid cartilage (Figure 1). The movements of the bilateral vocal folds were appropriate and symmetrical. Biopsy under general anaesthesia was performed and histopathology revealed a carcinoid tumour. Serum concentrations of tri-iodothyronine, thyroxine, thyroid-stimulating hormone, calcium and calcitonin and urine 5-hydroxyindole acetic acid (5-HIAA) levels were within normal ranges.

Computed tomography (CT) of the neck confirmed a small lesion in the right arytenoepiglottic fold but incidentally revealed a suspicious lymph node in level IV of the right neck. On ultrasonography, the lymph node was spherical, approximately 6 mm in diameter and heterogeneous with tiny calcifications (Figure 2). However, no significant tumour lesion was found in the thyroid gland. Echo-guided fine needle aspiration of the lymph node was performed, but cytological examination showed no malignancy.

Complete excision of the laryngeal tumour and adjacent corniculate and arytenoid cartilages, with a safety margin, was performed by use of a potassium titanyl phosphate (KTP) laser through a Lindholm laryngoscope. Selective neck dissection of levels II, III and IV of the right neck was also performed simultaneously.

Histopathology showed a typical carcinoid tumour composed of uniform polygonal cells with finely granular chromatin arranged in a trabecular pattern. Tumour cells were immunoreactive to cytokeratin, neuron-specific enolase and chromogranin (Figure 3). In the neck specimen, 20 lymph nodes were dissected, none of which contained tumour cells from the laryngeal carcinoid.

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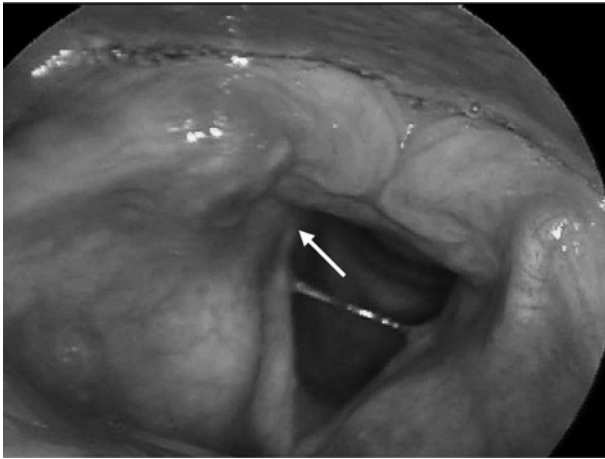


FIG. 1

Laryngoscopic view of a well demarcated, hyperaemic, lobulated, submucosal mass bulging at the superior surface of the right arytenoid cartilage (arrow).

However, one showed metastatic papillary carcinoma. Immunohistochemical staining was positive for thyroglobulin, suggestive of a papillary carcinoma of thyroid origin (Figure 4).

Despite the absence of a thyroid lesion detected on ultrasonography and CT scan, total thyroidectomy was performed two weeks later. No significant gross lesion was detected in the thyroid gland. Three tiny foci of papillary carcinoma were microscopically noted in the right lobe, the largest being 0.2×0.1 cm. Six months after thyroid surgery, the patient received radioiodine therapy. Two years later, he was alive, well and diseasefree.

Discussion

Typical carcinoid tumour of the larynx is extremely rare; only 16 cases (including this case) have been reported in the English literature.^{2,7}

Histologically, a typical carcinoid tumour is made up of sheets and nests of uniform cells with small, round, centrally placed nuclei in a poorly delineated, clear or slightly granular, eosinophilic cytoplasm. On immunohistochemical staining, the cells show positive immunoreactivity

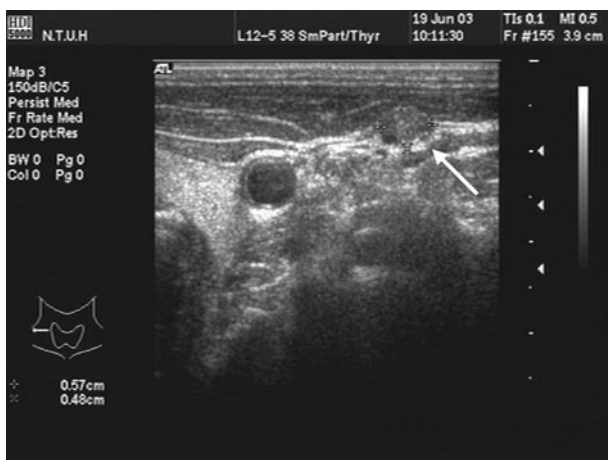


FIG. 2

Neck ultrasonography revealed a 0.57×0.48 cm, spherical, heterogeneous lymph node (arrow) at level IV of the right neck.

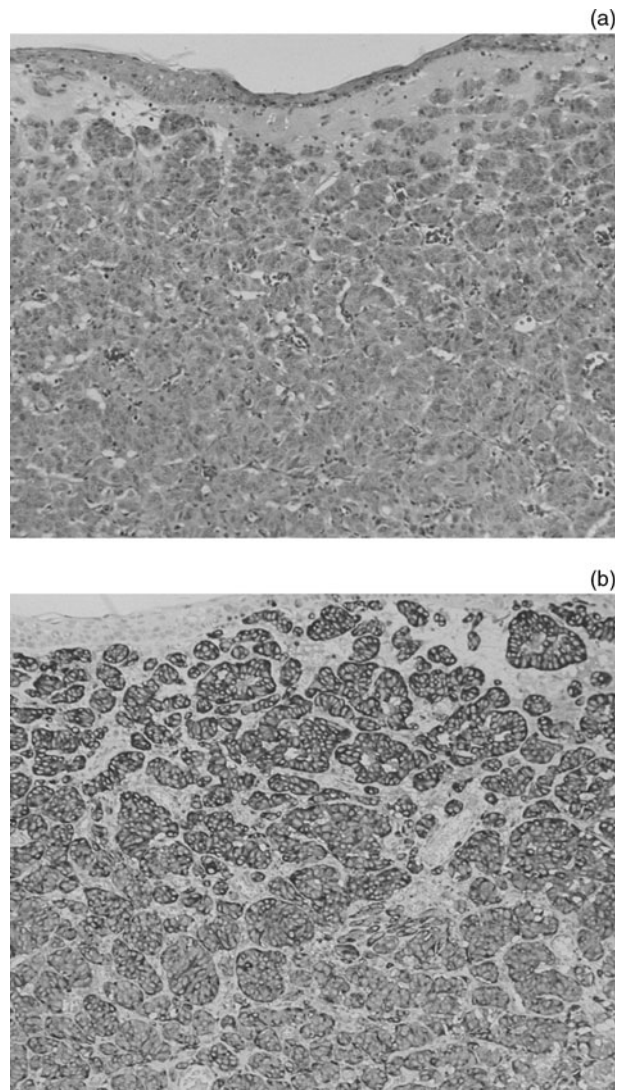


FIG. 3

Histopathological findings of typical carcinoid tumour of the larynx. (a) Tumour cells growing in a trabecular pattern, with a fine vascular stroma in the submucosal layer (H&E; $\times 33$ original magnification). (b) Tumour cells show strong cytoplasmic chromogranin staining (avidin-biotin-peroxidase complex (ABC) method; $\times 33$ original magnification).

for cytokeratin, neuron-specific enolase, calcitonin, somatostatin, carcinoembryonic antigen, chromogranin and serotonin. Compared with typical carcinoid, atypical carcinoid tumour shows more cytological atypia, greater mitotic activity and the presence of necrosis.^{1,2}

From previous experience, typical carcinoid tumour of the larynx tends to be localised. Metastasis (either nodal or distant) is rare at the time of diagnosis.^{2,7} Local excision without prophylactic lymph node dissection is adequate for treatment of this tumour, and results in long-term disease control. For preservation of the laryngeal framework, transoral endoscopic laser surgery is a promising approach for excision of benign laryngeal tumours or small, localised malignant tumours, and results in good local control and voice preservation.⁸

We originally planned to use a KTP laser via a laryngoscope to remove the laryngeal tumour, without neck dissection. However, the pre-operative CT scan and neck ultrasonography revealed a suspicious lymph node in the ipsilateral neck. This lymph node was small, and cytological

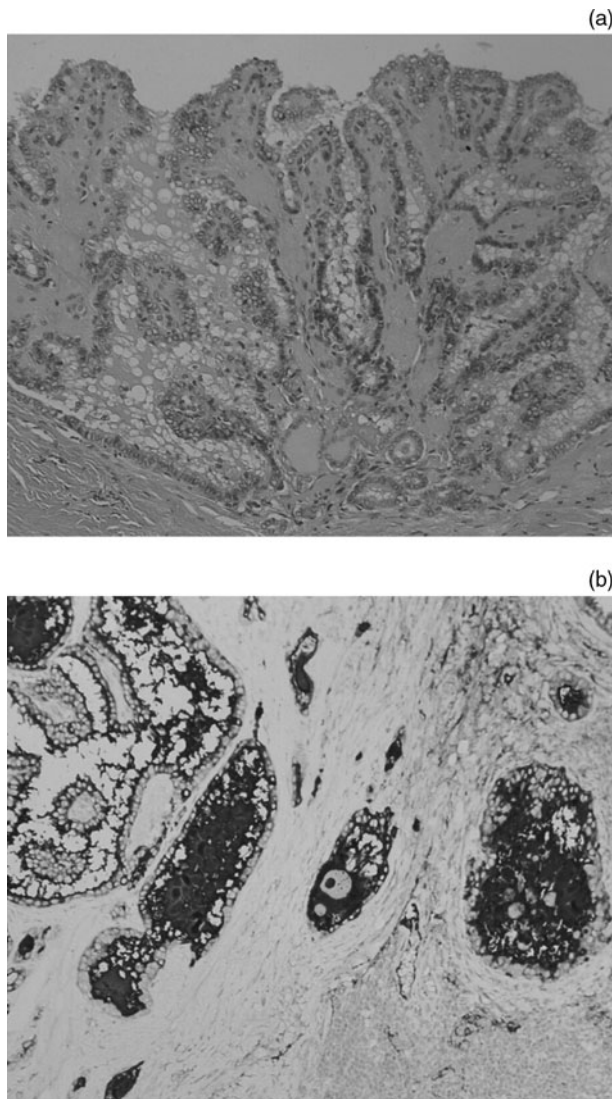


FIG. 4

(a) The cervical lymph node revealed a metastatic papillary carcinoma with complex, branching papillae and a central fibrovascular core (H&E, $\times 33$ original magnification). (b) Tumour cells demonstrated strong cytoplasmic staining for thyroglobulin (avidin-biotin-peroxidase complex (ABC) method; $\times 33$ original magnification).

examination did not reveal malignant cells. However, metastatic lymphadenopathy was still suspected because of the node's expansile shape (with a near-equal ratio of short to long axis), heterogeneous contents and lack of obvious hilum structure on ultrasonography. Therefore, excision of the laryngeal tumour via an endoscope was performed, with a selective neck dissection.

The histopathological examination of the laryngeal tumour showed a typical carcinoid tumour. However, the lymph node revealed by imaging studies was microscopically found to contain sheets of neoplastic cells with 'ground glass' nuclei and a papillary growth pattern, characteristics completely distinct from the carcinoid tumour cells. Metastatic thyroid papillary carcinoma was confirmed by thyroglobulin immunohistochemical staining.

It had been reported that unexpected pathological findings may be present in about 3 per cent of neck dissections undertaken for primary head and neck mucosal malignancies.⁴ Of these, metastatic papillary thyroid carcinoma is the most common incidental malignancy found. When

thyroid follicular or papillary carcinoma is discovered in cervical lymph nodes in the presence of a clinically and radiologically normal thyroid gland, some surgeons suggest observation alone. Their rationale is that such cases may represent so-called aberrant thyroid tissue in the cervical lymph nodes, and microscopic thyroid cancer may be stationary, without progression to clinically overt thyroid cancer. The overall prognosis is usually determined by the original index cancer, such as head and neck squamous cell carcinoma, and not by the occult thyroid cancer.^{4,9,10}

- **Typical carcinoid tumour of the larynx itself is extremely rare**
- **This paper describes a case with typical carcinoid tumour of the larynx and a simultaneous papillary thyroid carcinoma; this has not previously been reported in the literature**
- **Endoscopic excision of the laryngeal carcinoid tumour and total thyroidectomy with neck dissection and subsequent radioiodine therapy may have cured this patient**

However, in such cases, most surgeons suggest total thyroidectomy for eradication of all cancer foci, because primary tumours in the thyroid gland are usually multicentric and too tiny to be detected.^{11–15} Total thyroidectomy is not difficult and allows post-operative monitoring with thyroglobulin measurements and radioiodine scans.^{11–15} In our case, the neoplastic cells in the lymph node were demonstrated to be papillary carcinoma. Papillary thyroid microcarcinoma with lymph node metastasis may become the index cancer after excision of the curable laryngeal carcinoid tumour. Therefore, total thyroidectomy was performed. The final histopathological examination of the entire thyroid gland confirmed multifocal papillary carcinoma, each focus being less than 0.2 cm in diameter.

Conclusion

Typical carcinoid tumour of the larynx appearing simultaneously with papillary carcinoma of the thyroid gland has not previously been reported. Endoscopic laser excision of the laryngeal tumour and total thyroidectomy with neck dissection and subsequent radioiodine therapy probably cured this patient.

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