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Invasion of the recurrent laryngeal nerve by adenoid cystic carcinoma. An unusual cause of true vocal fold paralysis

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Abstract
True vocal fold paralysis and goitre are both common problems encountered in ENT practice. Their co-existence, however, should arouse suspicion of the presence of malignant thyroid disease. A rare case of true vocal fold paralysis caused by a clinically occult subglottic adenoid cystic carcinoma, in a 72-year-old, is described. The existence of multinodular goitre in this patient was co-incidental and confounded the diagnostic process.

Key words: Vocal fold paralysis; Thyroid neoplasms; Carcinoma, adenoid cystic

Case report

A 72-year-old lady presented to the clinic with a six-week history of persistent hoarseness and mild neck pain which localized to the cricoid level in the midline. Examination revealed a left true vocal fold palsy (TVFP) with a multinodular goitre (MNG) which extended retro-sternally. There was no cervical lymphadenopathy, dyspnoea or stridor. Ultrasound scan of the thyroid gland confirmed a MNG but failed to detect any other mass. Chest X-ray and barium swallow were normal and fine needle aspiration cytology of the thyroid was consistent with MNG. Computed tomography (CT) scan of the neck demonstrated an abnormal area of soft tissue medial to the left lobe of the thyroid causing some indentation of the trachea (Figure 1), as well as clearly demonstrating the MNG (Figure 2). Rigid endoscopy and biopsy failed to show a lesion and subsequent CT-guided biopsy failed to show any pathological lesion. The goitre increased in size over the following two months and therefore a diagnostic left thyroid lobectomy and repeat endoscopy was undertaken. At thyroid exploration, a tumour was found deep to the left cricopharyngeus muscle extending along the recurrent laryngeal nerve (RLN) in the tracheo-oesopha-

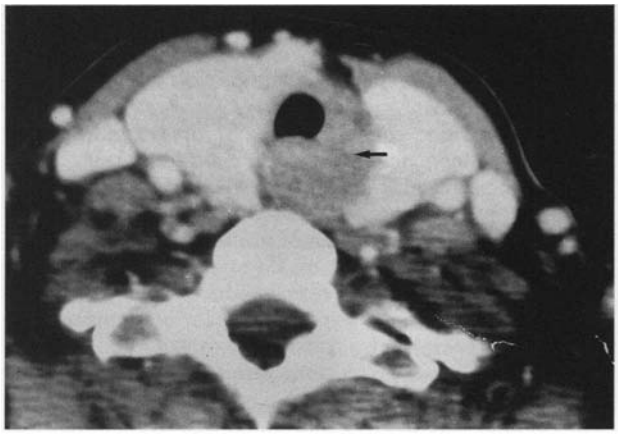

Fig. 1
Axial contrast enhanced CT scan through the trachea reveals enlargement of both lobes of the thyroid with an added soft tissue mass of intermediate attenuation seen lying between the trachea and the medial aspect of the left lobe of the thyroid (arrow). The oesophagus cannot be distinguished separately from this mass. The mass is causing some posterior indentation of the tracheal lumen but this can be seen in a normal patient at this level.

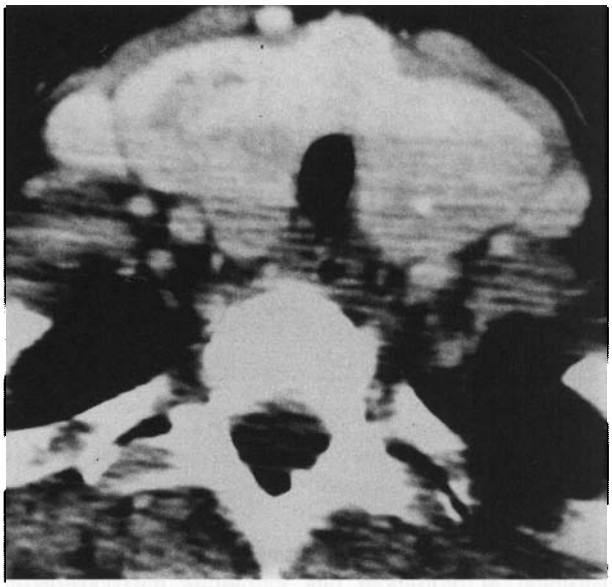

Fig. 2
Axial CT scan following intravenous contrast through the root of the neck reveals marked enlargement of both lobes of the thyroid with areas of decreased attenuation in both lobes consistent with a multi-nodular goitre. This is causing some compression of the trachea.

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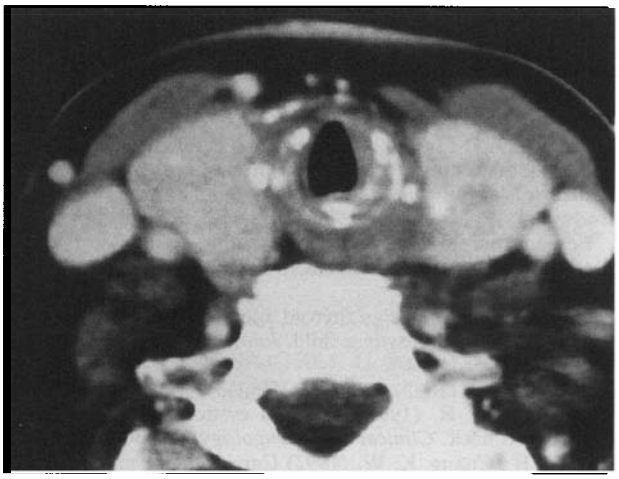

FIG. 3a

Axial CT scan following i.v. contrast through the level of the cricoid cartilage. At this time there is symmetric ossification of the cricoid cartilage. Some added soft tissue is apparent lying between the cricoid cartilage on the left and the left lobe of the thyroid.

geal groove into the thyroid gland. There was obvious macroscopic invasion of the nerve. A subglottic tumour was endoscopically visible but it did not breach the mucosa. The post-operative period was complicated by a brief episode of stridor which resolved on dexamethasone. Histological examination of the thyroid gland showed MNG invaded posteriorly by adenoid cystic carcinoma (ACC). Further CT scan one week later showed added soft tissue in the subglottic region with sclerosis of the left side of the cricoid cartilage which had not existed on the earlier CT scan of the neck (Figure 3a and 3b). As the tumour was

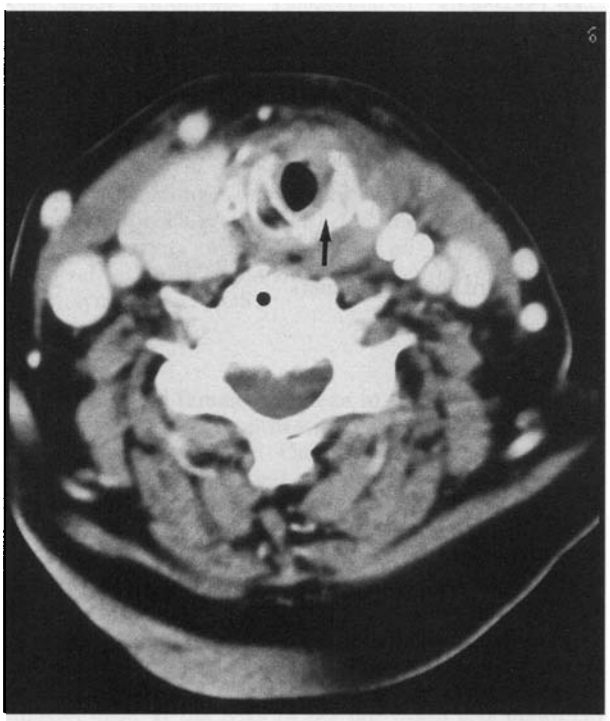

FIG. 3b

Axial CT scan following i.v. contrast in the same patient two months later (post-left thyroid lobectomy). This scan demonstrates increased sclerosis of the left side of the cricoid cartilage (arrow). This can be an indicator of tumour invasion and this was subsequently confirmed by histological examination.

incompletely excised, a completion total laryngectomy was undertaken. Histological examination showed apparent complete resection of a well-differentiated ACC which primarily involved the subglottis. The tumour was invading both the left RLN at the upper thyroid pole and also the cricoid cartilage on the left (previously indicated on CT scan – see figure 3b).

Discussion

Adenoid cystic carcinoma of the subglottis

Despite the benign-sounding synonym of 'cylindroma', ACC is always malignant and in 1.5 per cent of these tumours the primary site is the larynx. The tumour arises from the seromucinous glands in the larynx (Tewfik *et al.*, 1983), and the distribution of these glands is reflected in the relative frequency of ACC in the subsites of the larynx. In one case series, out of 33 laryngeal ACCs, 18 were subglottic, 11 were supraglottic and four cases were either glottic or trans-glottic (Batsakis *et al.*, 1992). Subglottic lesions usually present with stridor; other symptoms being neck pain (due to neural involvement) and cough. Hoarseness is not a common presenting feature but may occur if a true vocal fold is fixed by crico-arytenoid muscle invasion, or as in this case, by RLN involvement and TVFP. TVFP due to RLN invasion by laryngeal ACC is previously unreported. One similar case is of a tracheal ACC invading the RLN and thyroid gland resulting in goitre being the presenting symptom (Zirkin and Tovi, 1984).

Characteristics of both the anatomical site and tumour behaviour determine the pattern of spread of ACC in this region. Subglottic anatomy provides little defence against local tumour spread. The inferior margin of the thyroid cartilage extends 10 mm below the level of the true vocal folds antero-laterally, but is somewhat deficient posteriorly, with only 3–4 mm of inferior extension. The mucosa below the lower border of thyroid cartilage directly abuts the cricothyroid membrane or cricoid space and hence provides a direct passage for extra-laryngeal invasion. Consequently, subglottic malignancies tend to present late with 45 per cent being T4 at presentation (Shaha and Shah, 1982). This lack of a natural barrier to local invasion of subglottic cancer is exacerbated by the fact that ACC in this region commonly spreads extensively along perineural planes and has been shown to frequently invade cartilage directly (Maziak *et al.*, 1996). The tumour in the case described here was staged as T4 and demonstrated direct invasion of both cartilage and perineural tissues. It also had undergone early submucosal spread (common in ACC) with little formation of an exophytic mass, and this had resulted in the negative endoscopic examination. CT scanning has however been shown to be effective at detecting the diffuse morphology of ACC in this anatomical region (Na *et al.*, 1995). Early lymphatic spread to regional lymph nodes is uncommon (Tewfik *et al.*, 1983), however death is typically preceded by late neck nodal recurrence (23 per cent at 15 years) as well as late local recurrence (100 per cent at 30 years) and a high rate of late pulmonary metastases (Jones *et al.*, 1997). Treatment is by primary wide local excision (total laryngectomy, in the case of subglottic ACC) with adjuvant external beam radiotherapy to the primary site. The overall tumour-specific survival rate for all ACCs of the head and neck is 40 per cent at 20 years (Jones *et al.*, 1997). The corresponding figure for the rare cases of ACC of the subglottis is unknown but prognosis appears to be similarly dismal regardless of treatment (Donovan and Conley, 1983).

Coexistence of goitre and true vocal fold paralysis

The prevalence of goitres in the United Kingdom is approximately one in 10. Between 1.4 per cent (Franklyn *et al.*, 1993) and 7.5 per cent (Koh and Chang, 1992) of all MNGs harbour malignancy – most commonly papillary. TVFP is occasionally caused by an advanced thyroid malignancy involving the RLN and so any association of goitre and RLN palsy should therefore prompt further investigation such as CT scanning. Rarely, a benign goitre can cause TVFP which may be uni- or bi-lateral. Examples of such benign pathologies include hyperplasia (Godwin *et al.*, 1991), adenoma (Habashi, 1991), simple cyst (Quayle and Talbot, 1987), tuberculous abscess (Emery, 1980), Reidel's thyroiditis and Graves' disease (Falk and McCaffrey, 1995). Of these cases, 89 per cent will recover normal vocal fold mobility with thyroidectomy (Rowe-Jones *et al.*, 1993). In the case described here, thyroid lobectomy was justified because the enlarging goitre was the only apparent cause for the TVFP and therefore its removal could have led to RLN recovery. In this instance however, thyroid lobectomy served only to unmask occult malignancy in an adjacent organ.

Summary

MNG is an important finding in a case of TVFP. The RLNs are susceptible to being involved in the spread of laryngeal ACC. It is for this reason that this patient's symptoms of neck pain and TVFP were key indicators for the presence of an ACC of her larynx. Extensive subglottic ACC, with invasion of the laryngeal framework and adjacent structures, may remain entirely submucosal and elude diagnosis using rigid endoscopy. Contrast-enhanced CT is therefore of particular value in this situation. Diagnostic thyroidectomy is justifiable when the only causal factor for TVFP is a goitre.

References

Batsakis, J. G., Luna, M. A., El-Naggar, A. K. (1992) Nonsquamous carcinomas of the larynx. *Annals of Otology, Rhinology and Laryngology* **101:** 1024–1026.

Donovan, D. T., Conley, J. (1983) Adenoid cystic carcinoma of the subglottic region. *Annals of Otology, Rhinology and Laryngology* **92(5 Pt 1):** 491–495.

Emery, P. (1980) Tuberculous abscess of the thyroid with recurrent laryngeal nerve palsy: case report and review of the literature. *Journal of Laryngology and Otology* **94:** 553–558.

Falk, S. A., McCaffrey, T. V. (1995) Management of the recurrent laryngeal nerve in suspected and proven thyroid cancer. *Otolaryngology – Head and Neck Surgery* **113:** 42–48.

Franklyn, J. A., Daykin, J., Young, J., Oates, G. D., Sheppard, M. C. (1993) Fine needle aspiration cytology in diffuse or multinodular goitre compared with solitary thyroid nodules. *British Medical Journal* **307:** 240.

Godwin, J. E., Miller, K. S., Hoang, K. G., Sahn, S. A. (1991) Benign thyroid hyperplasia presenting as bilateral vocal cord paralysis. Complete remission following surgery. *Chest* **99:** 1029–1030.

Habashi, S. (1991) Benign thyroid adenoma causing recurrent laryngeal nerve palsy in a child. *Journal of Laryngology and Otology* **105:** 141–142.

Jones, A. S., Hamilton, J. W., Rowley, H., Husbard, D., Helliwell, T. R. (1997) Adenoid cystic carcinoma of the head and neck. *Clinical Otolaryngology* **22:** 434–443.

Koh, K. B., Chang, K. W. (1992) Carcinoma in multinodular goitre. *British Journal of Surgery* **79:** 266–267.

Maziak, D. E., Todd, T. R., Keshavjee, S. H., Winton, T. L., Van Nostrand, P., Pearson, F. G. (1996) Adenoid cystic carcinoma of the airway: 32-year experience. *Journal of Thoracic and Cardiovascular Surgery* **112:** 1522–1531. (Discussion 1531–2).

Na, D. G., Han, M. H., Kim, K. H., Chang, K. H., Yeon, K. M. (1995) Primary adenoid cystic carcinoma of the cervical trachea mimicking thyroid tumour: CT evaluation. *Journal of Computed Assisted Tomography* **19:** 559–563.

Quayle, A. R., Talbot, C. H. (1987) Simple thyroid cyst: cause of acute bilateral recurrent laryngeal nerve palsy [letter]. *British Medical Journal* (Clinical Research Edition) **294:** 1487.

Rowe-Jones, J. M., Rosswick, R. P., Leighton, S. E. (1993) Benign thyriod disease and vocal cord palsy. *Annals of the Royal College of Surgeons of England* **75:** 241–244.

Shaha, A. R., Shah, J. P. (1982) Carcinoma of the subglottic larynx. *American Journal of Surgery* **144:** 456–458.

Tewfik, T. L., Novick, W. H., Schipper, H. M. (1983) Adenoid cystic carcinoma of the larynx. *Journal of Otolaryngology* **12:** 151–154.

Zirkin, H. J., Tovi, F. (1984) Tracheal carcinoma presenting as a thyroid tumour. *Journal of Surgical Oncology* **26:** 268–271.

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