A Rare Instance of Co-existing Malignancy in Ectopic and Orthotopic Thyroid Gland

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ABSTRACT

Ectopic thyroid tissue can be found anywhere in the midline from the base of the tongue to mediastinum. Ectopic thyroid tissue may also be involved in the same pathological processes like tumors, inflammation and hyperplasia as normal, orthotopic thyroid gland. Besides, the appearance of such tissue in rare locations may result in diagnostic and therapeutic dilemmas. We report an extremely rare case of simultaneously occurring papillary carcinoma in ectopic subglottic thyroid and orthotopic thyroid gland in a young Asian male. The patient presented with symptoms completely unrelated to thyroid disorder, and went through an eventful journey before being correctly diagnosed and properly treated.

Keywords: Thyroid carcinoma, ectopic thyroid, orthotopic thyroid.

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INTRODUCTION

Ectopy is the most common developmental anomaly of the thyroid gland (1). Developmental defects in the early thyroid gland embryogenesis may result in ectopia, at any place during its path of descent from the primitive foregut floor to the ultimate pre-tracheal position (2). It is usually found in the lingual, thyroglossal and laryngotracheal sites, with a reported incidence of 1 in 300,000 (3). Although most cases of ectopic thyroid are asymptomatic, any disease affecting the thyroid may potentially involve the ectopic tissue, including malignancies. The probability of carcinoma arising in ectopic thyroid is reported to be less than 1% (4). However, the appearance of such tissue in rare locations may lead to diagnostic and therapeutic dilemmas (3).

CASE REPORT

Our patient is a 22 years old Asian male who was experiencing persistent and progressive hoarseness of voice, exertional dyspnoea & occasional haemoptysis for two years. There was no history of fever, pain, palpitation, insomnia, visible neck swelling, weight gain or loss, or any other abnormality.

His initial investigations showed normal thyroid hormone status and a mildly enlarged thyroid gland with non-homogeneous parenchyma on high resolution ultrasound (figure 1). Ultrasound guided fine needle aspiration (FNA) from right lobe of thyroid gland revealed benign nodular goiter, Bethesda category II.

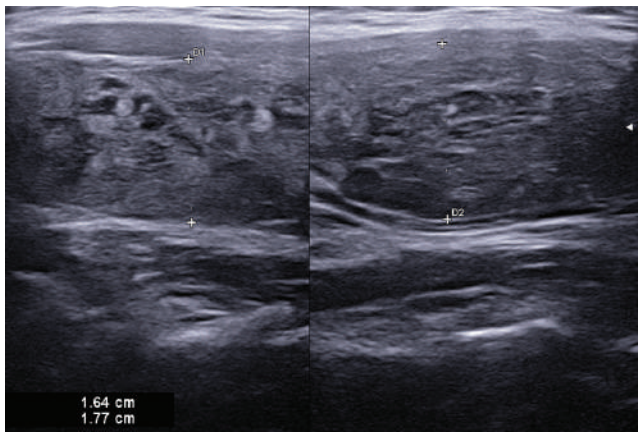

Figure 1: High resolution ultrasound of thyroid gland showing mild thyromegaly with non-homogenous parenchyma giving early nodular change.

Contrast computed tomography (CT) of neck showed a subglottic mass causing narrowing of subglottic airway (figure 2).

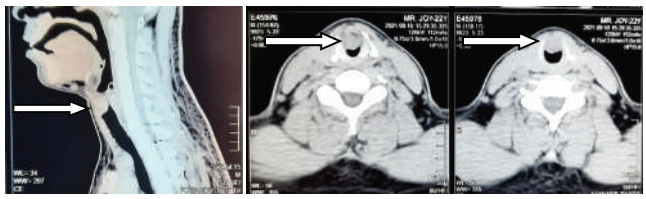

Figure 2: Contrast CT neck showing a moderate homogeneously enhancing well circumscribed soft tissue density lesion (white arrow) measuring about (18 x 20 mm) in subglottic region beneath the anterior commissure

of vocal cord causing partial airway obstruction. There was no evidence of erosion or destruction of thyroid cartilage.

Laryngoscopy was performed and a haemorrhagic polyp was found in subglottis that bleeds on touch (figure 3).

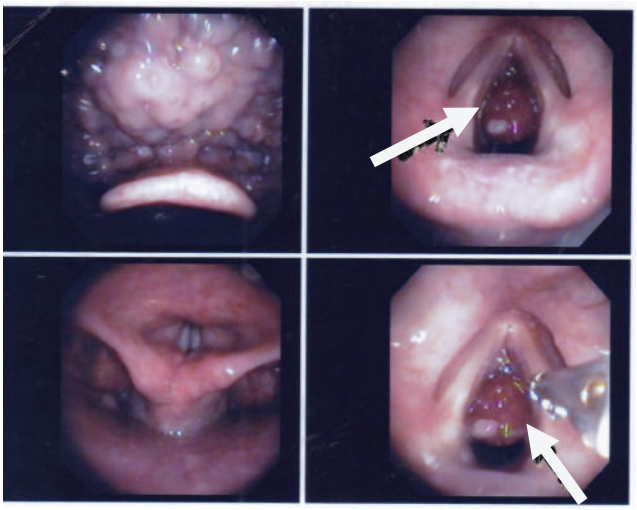

Figure 3: Laryngoscopic image showing a subglottic mass suggesting hemorrhagic polyp (white arrow).

Based on these findings his differential diagnosis was subglottic hemangioma or carcinoma. But histopathology from CT guided FNA from the intra tracheal growth suggested papillary carcinoma, while laryngoscopic biopsy reported ectopic thyroid tissue with chronic inflammatory cells and no malignancy. At this point the patient decided to go to abroad for surgical treatment. There, his pre-operative assessment again showed a normal blood picture with non-diabetic and euthyroid status. CT scan of neck commented on the well-defined subglottic mass to be relatively hypodense to thyroid tissue, and intensely vascular on correlative ultrasound, so a vascular lesion like hemangioma could not be excluded from radiological point of view. A necrotic lymph node was also seen at right level IIa. Thyroid gland showed diffuse heterogeneous echotexture with no focal lesion.

He underwent CO2 laser assisted micro laryngoscopy and complete excision of the subglottic lesion. Histopathological examination of the specimen confirmed papillary carcinoma. After that, completion thyroidectomy was done with right selective neck dissection. Histopathology of the thyroid gland revealed classic papillary microcarcinoma in right lobe, with a background of lymphocytic thyroiditis, having no lymphovascular invasion or extrathyroidal extension, and no lymph node involvement. So, the final diagnosis was papillary microcarcinoma of thyroid gland with papillary carcinoma in subglottic ectopic thyroid.

Then the patient came back and was referred to our institute for post-operative radio-iodine therapy. He was off thyroxin for four months since surgery and we found S. TSH > 100 µIU/ml, S. Thyroglobulin 0.17 ng/ml. Ultrasound of neck and thyroid scan showed normal post thyroidectomy state. We administered 75 mCi oral radio-iodine to him following all radiation safety protocol. His post therapy scan revealed a focal uptake at pelvis (figure 4). We also performed a bone scan which showed no suspicious osteoblastic lesion.

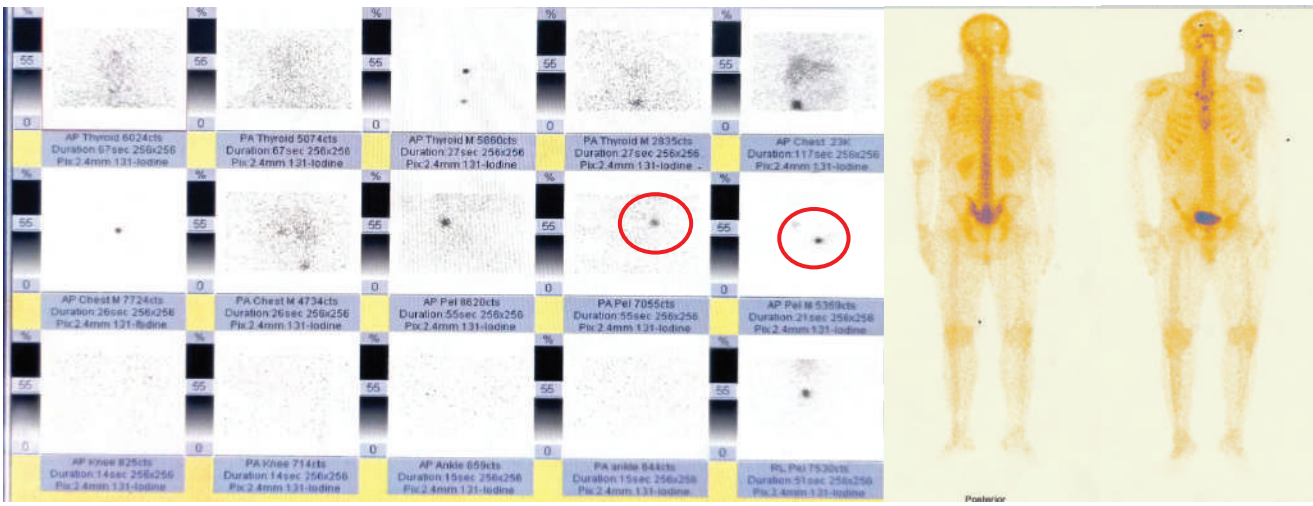

Figure 4: Post therapy whole body scan showing a faint uptake at pelvis (red circle), and on right hand side, Tc-99m bone scintigraphy image revealing normal radiotracer distribution.

Following an uneventful post-therapy recovery, the patient remains asymptomatic till date and following our regular follow up schedule. All of his parameters are within normal limit with no significant complaint.

DISCUSSION

Though majority of thyroid cancers (about 99%) develop within the eutopic thyroid gland itself, rarely a carcinoma can arise in ectopic thyroid tissue (3). Most common ectopic thyroid malignancies are papillary thyroid carcinomas originating from thyroglossal duct cysts and lingual thyroid. Thyroglossal duct cyst carcinomas can appear as an isolated lesion, or with a co-existing malignancy in orthotopic thyroid gland (1).

Intratracheal ectopic thyroid tissue accounts for about 6-7% of all primary endotracheal tumors. It can be found anywhere from glottis to tracheal bifurcation, but most commonly on the lateral subglottic and upper tracheal wall as a submucosal mass (5). The patient in this case also presented with a subglottic mass. Ziemssen described the first case of intratracheal goiter in 1875, and Byrd et al. reported that from 1966 to 2003 there have been only 13 well-documented cases of intratracheal ectopic thyroid in the literature, only two of those malignant. Malignant change in intratracheal ETT is most commonly papillary thyroid carcinoma, as in our patient (6).

Patients may remain symptomless, but when symptomatic ectopic intra-tracheal thyroid causes breathing difficulty, cough, stridor, and dysphagia. Findings suspicious of malignancy include multiple nodules, ulceration, and bleeding (5). In a symptomatic case, complete ear, nose and throat evaluation should be performed, as well as thyroid assessment. Other investigations including laryngoscopy, CT and magnetic resonance (MR) studies must be included. For detecting functional thyroid tissue in ectopic location radionuclide studies are highly sensitive and specific (7). Thyroid scintigraphy using Tc-99m pertechnetate can effectively assess the size, distribution, and functional status of ectopic intra-tracheal thyroid. Another valuable nuclear medicine test in this regard is fusion imaging techniques like single photon emission tomography-computed tomography (SPECT-CT). Early application of these techniques could have saved our patient from undergoing a prolonged and complicated path for a proper diagnosis.

Management of ectopic intra-tracheal thyroid depends on patient's age, size of the lesion, symptoms. thyroid function status, and histological findings. Main treatment options include surgical excision, radioiodine ablation, and thyroid suppression therapy (8).

CONCLUSION

Co-existence of carcinoma in both ectopic and orthotopic thyroid gland is a very rare occurrence. Furthermore, the rare location of ectopic tissue in this case has also increased the magnitude of its uniqueness. The lesson learnt from this unique case is that presentation of a thyroid pathology can be very diverse, and hence physicians have to walk extra miles to explore even the remotest possibilities before a correct diagnosis. Successful management has been made possible by the combined effort of multiple disciplines including surgeons, pathologists and nuclear medicine clinicians.

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