



Letter to the editor

Lingual ectopic papillary thyroid carcinoma: Two case reports and review of the literature



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ABSTRACT

Ectopic thyroid occurs when it is not located on the normal thyroid compartment. While 90% of the ectopic thyroids were located at the base of the tongue, only 1% were lingual thyroid carcinoma (LTC). Only 56 LTC cases have been reported so far. Here we reported two cases of LTC. Patient 1 was a 47-year-old female with LTC and co-current sub-hyoid ectopic thyroid. She experienced major hematemesis and dyspnea requiring emergent tracheotomy. Patient 2 was a 61-year old female who was presented with LTC with multiple lymph node metastasis and bilateral lung metastasis. Both of the patients' lingual masses were removed via trans-submaxillary excisions. Pathology revealed ectopic papillary thyroid carcinoma. Then they were treated with radio-active iodine (RAI). These patients had full recovery and there were no complications. A review of literature was also presented.

Introduction

Ectopic thyroid occurs when it is not located on the normal thyroid compartment. The majority of ectopic thyroid develops any point from the foramen cecum in the tongue base to the mediastinum, along the embryologic descent of the thyroid anlage [1]. 90% of the ectopic thyroids were located at the base of the tongue and only 1% were lingual thyroid carcinoma (LTC) [2].

Patient 1

A 47-year-old female was first presented with a lingual mass in July 2010. Computed tomography (CT) scan demonstrated a 4.5 * 4.5 cm lingual mass, and a submandibular mass of 2.0 * 1.8 cm with the absence of thyroid tissue before the trachea. She was diagnosed with lingual ectopic thyroid and was administered with L-thyroxine tablets. She had a history of thyroglossic cystectomy 20 years ago.

However, in recent six months she gradually developed dysphagia, and sensation of foreign body. On 17th February 2017, the patient was presented with acute dyspnea. On her way to the emergency, she coughed out a mass with hematemesis of 1000 ml which relieved her dyspnea. Tracheotomy was performed immediately in the local hospital. Lab results showed her hemoglobin was 60 g/L, and she was transferred with 2 units of red blood cells. The mass which she coughed out was sent for pathology analysis and revealed to be papillary thyroid carcinoma (PTC).

On 11th April 2017, she underwent partial glossectomy by trans-submaxillary excision. Three months later, the patient was treated with 50 mCi ¹³¹Iodine. The scan after treatment showed no residue of lingual thyroid carcinoma, and there was a 15 * 6 mm mass in the anterior

neck, indicating sub-hyoid ectopic thyroid tissue. She was then started TSH-suppression therapy. The 6-months post-iodine treatment CT scan and ultrasound showed no residue thyroid tissue. The patient remained disease-free on her last follow-up in November 2018.

Patient 2

A 61-year old female was presented with hoarseness in 2006 and a mass on her tongue base was subsequently discovered. No further pathology assessment was performed at the time. In October 2017, she was presented with short of breath and PET-CT scan showed lingual malignancy, with bilateral neck and lung metastasis. However, tongue biopsy revealed hyperplastic fibrous tissue with necrosis and inflammatory; and ultrasound guided-fine needle aspiration (FNA) of the left neck showed no evidence of malignancy (Tg measurement was not available at our institute for FNA). She also had evaluated TSH level (6.7576 mIU/L) with normal T3 and T4.

On January 2018, the lingual mass was removed via a trans-submaxillary excision. Pathology revealed ectopic PTC (Fig. 1). The patient was then treated with 200 mCi ¹³¹Iodine. Residue lingual thyroid, bilateral cervical lymph nodes and upper mediastinal lymph node and bilateral lung nodules were identified on the post-treatment scan (Fig. 1). Another session of radio-active iodine (RAI) treatment was scheduled for her in six months.

Discussion

We presented two typical cases of LTC. The first case was characterized by LTC with co-current sub-hyoid ectopic thyroid. She also had a history of thyroglossic cystectomy, indicating a series of diseases

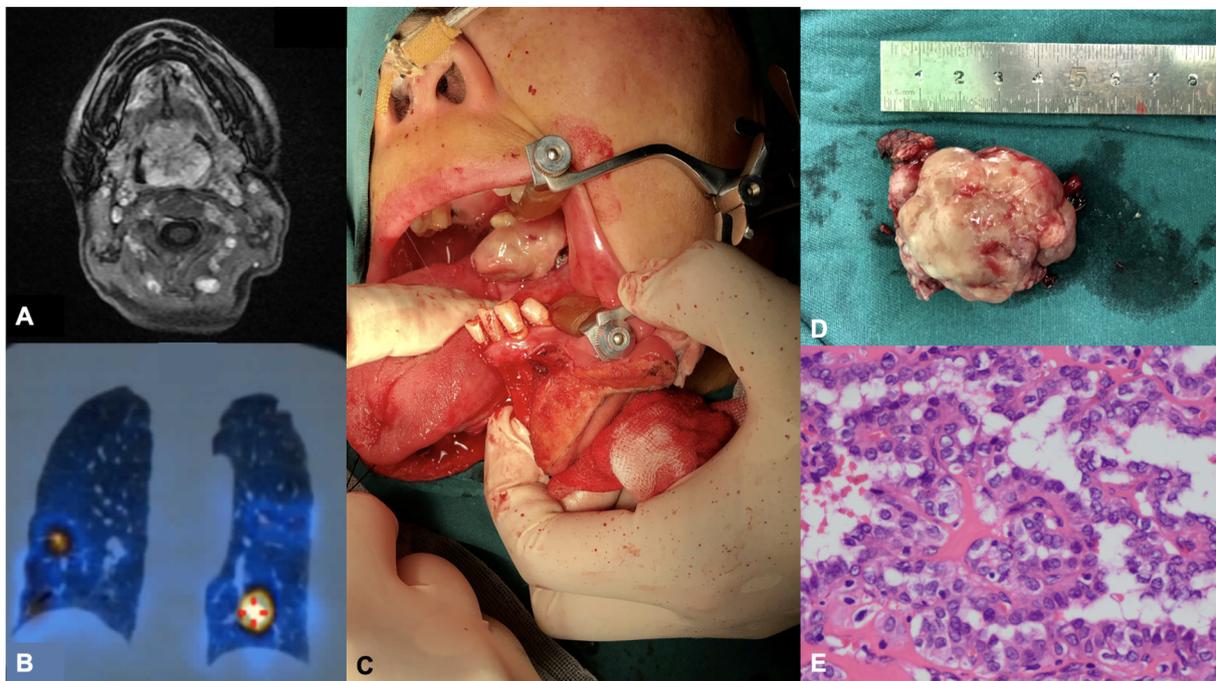


Fig. 1. (A) Pre-operative magnetic resonance imaging (MRI) of Case 2 showed an irregular 38 * 32 mm mass of the tongue. (B) Post-radio-active iodine treatment scan of Case 2 showed bilateral lung metastasis. (C) This photo shows that the position of the tumor was at the base of the tongue and could be exposed via mandibulotomy; 1D: The size of the tumor was 4.5 cm × 3.0 cm; 1E: Final pathology on the HE section (× 400) revealed classical papillary thyroid carcinoma.

caused by the failure to decent of the thyroid anlage. To our knowledge, it was also the first case reported to have LTC, co-current sub-hyoid ectopic thyroid, and thyroglossic cyst in a single patient. Another highlight of the first case was that, the patient had major hematemeses and dyspnea requiring emergent tracheotomy caused by LTC. Although hematemeses was not rare in LTC cases, our case showed that the sudden rupture of the LTC would cause major bleeding and emergent tracheotomy was needed to ensure safe airway.

The second case was LTC with multiple lymph node metastasis and lung metastasis. This case represented the 5th cases of LTC with lung metastasis reported [3–6]. The patient was not properly diagnosed and treated when she first found the tongue mass 11 years ago due to the rare nature of the disease.

Then, we searched Pubmed with the key words: “lingual”, “tongue”, “thyroid”, “ectopic”, “cancer”, “carcinoma” and “malignancy(ies)” for literature review. We identified 58 cases of LTC in total from 1910 to 2018, including the current two cases. The majority of patients complained of dysphagia, change of voice or oral bleeding with absent of orthotopic thyroid. And more than half of the patients with thyroid function tests had hypothyroidism. Some authors suggested that orthotopic thyroid gland might cause hypothyroidism [7,8].

Only 22 cases provided detailed information of treatment and were included into further analysis (Table 1). All patients had surgical excision of the lingual lesion except for Kao et al. treated the patient with total thyroidectomy and RAI [9]. 13 out of 21 cases had trans-oral

surgery, while the rest had trans-cervical surgery. More specifically, trans-hyoid approach, pharyngotomy, trans-mandibular approach, split tongue in the midline and direct resection trans-orally were used. The choice of surgical approach depended on the location, size of the tumor as well as the experience of the surgeons.

Neck dissection was recommended for LTC with cervical lymph node metastasis, indicating the importance of routine assessment of cervical lymph nodes. Interestingly, more than half of the cases reported to perform prophylactic tracheotomy, because of the edema of the airway; and all cases had uneventful post-operative course. In Stoke et. al.’s case without tracheotomy, the patient developed airway edema and required urgent reintubation [10]. Therefore, we recommended routine prophylactic tracheotomy for LTC patients. Remarkably, the treatment strategies of LTC had involved overtime. While surgery/radiation was commonly used in earlier cases [3,4], it had been replaced by surgery/RAI in more recent cases. Up to 16 patients had RAI in the 22 cases. In patients without RAI, one case was medullary carcinoma, and the others were not metastatic disease.

In conclusion, we added two LTC cases with typical presentations and clinical courses to the literature. Although LTC is rare, it should be taken into considerations in the differential diagnosis of lingual mass. The management of the disease, however, requires a multi-disciplinary approach.

Table 1
22 lingual thyroid carcinoma cases with detailed treatment information.

Year	Author	Sex	Age	Orthotopic thyroid	Thyroid function	Pathology	Metastasis	Surgery/ Radiation	Neck dissection	Tracheotomy	RAI
1971	Potdar [11]	M	12	No	Normal	Follicular	No	Transoral, split tongue in the midline	No	Yes	Yes
1979	Kamat [12]	F	40	NR	NR	Follicular	No	Transoral, split tongue in the midline	No	Yes	Yes
1979	Singh [13]	F	28	Yes	NR	Papillary	No	Trans-cervical, trans-hyoid	No	No	No
1980	Goode [14]	F	40	Yes	Normal	Follicular	No	Transoral	No	No	No
1992	Diaz-Arias [15]	F	23	No	Hypothyroidism	Follicular	No	Transoral	No	No	No
1997	Winslow [16]	M	23	No	Hypothyroidism	Papillary [follicular variant]	Cervical LN	Transoral, split tongue in the midline	Yes	Yes	Yes
2001	Massine [17]	M	57	No	Hypothyroidism	Papillary	NR	Transoral, trans-madibular*	Yes	Yes	Yes [#]
2002	Kao [9]	M	70	Yes	NR	Papillary	Cervical LN	External beam radiation	Yes	No	Yes
2002	Goldstein [18]	F	48	No	NR	Papillary	No	Trans-cervical, trans-hyoid	No	Yes	Yes
2003	Perez [19]	M	28	No	Hypothyroidism	Papillary	No	Transoral, split tongue in the midline	No	No	Yes
2005	Falvo [20]	F	30	Yes	Normal	Papillary [sclerosing]	Cervical LN	Trans-cervical, trans-hyoid	No	No	Yes
2007	Addams-Williams [21]	F	50	No	Normal	Papillary [follicular variant]	No	Trans-cervical, pharyngotomy	No	Yes	Yes
2007	Kennedy [22]	F	25	No	Hypothyroidism	Papillary	Cervical LN	Trans-cervical, trans-hyoid	Yes	Yes	Yes
2008	Yaday [23]	F	45	No	NR	Medullary	No	Transoral, trans-madibular	No	NR	No
2009	Raju [6]	F	45	NR	Normal	Papillary	No	Trans-cervical, pharyngotomy	No	Yes	No
2013	Chen [24]	M	42	Yes	Hypothyroidism	Papillary [follicular variant]	No	Trans-cervical, pharyngotomy	No	Yes	No
2017	Sturmiolo [11]	F	63	No	Normal	Follicular	Cervical LN	Transoral	Yes	No	Yes
2018	Stoke [10]	F	84	Yes	NR	Papillary	Cervical LN	Trans-cervical, trans-hyoid	Yes	No	Yes
2018	Mogi [25]	M	40	No	Hypothyroidism	Papillary	Cervical LN	Transoral	Yes	Yes	Yes
2018	Thakur [26]	F	37	No	Hypothyroidism	Hürthle	No	Transoral, split tongue in the midline	No	Yes	Yes
/	Huang	F	47	No	NR	Papillary [follicular variant]	No	Transoral, trans-madibular	No	Yes	Yes
/	Huang	F	61	No	Hypothyroidism	Papillary	Lung, cervical and mediastinal LN	Transoral, trans-madibular	No	Yes	Yes

M: male, F: female, NR: not reported, LN: lymph node, RAI: radio-active iodine.

* Tongue SCC was identified at the same time, and the total tongue was resected with flap reconstruction.

Pre-operative RAI was delivered because the patient refused surgery at first.

Conflict of interest statement

None declared.

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