

Removal of Ectopic Lingual Thyroid : A Case Report

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We report a rare case of ectopic lingual thyroid with a risk of strangulation. A 32-year-old female patient had a lingual tumor and computed tomography (CT) scan revealed a rounded mass with calcification, occupying the pharyngeal space. No thyroid gland was seen in the normal pretracheal position. ^{99m}Tc sintigraphy showed high uptake only in the sublingual region. In ²⁰¹Tl sintigraphy, wash out was poor which might have further complicated the malignant tumor. The patient was also found to suffer from subclinical hypothyroidism. We underwent lingual thyroidectomy using an external cervical approach. The patient had no apparent complication resulting from surgery and was discharged after two weeks.

Key word: ectopic lingual thyroid, ^{99m}Tc sintigraphy

INTRODUCTION

During the development of the thyroid gland during embryogenesis, the glandula thyroidea descends from the foramen cecum to the pretracheal portion [1]. Ectopic thyroid is an anomaly that is caused by undescendence of the embryonic thyroid gland or an aberration of the glandula thyroidea into the ductus thyroglossus [2]. Most patients live to death without any symptoms such as dyspnea or dysphagia [3]. Here we report a rare case which needed

the surgical removal of the lingual thyroid as the ectopic thyroid enlarged to occupy the pharyngeal space during hypothyroidism.

CASE REPORT

In August, 2009, a 32-year-old female presented a cough to a local practitioner which was diagnosed as a tumor located in the epiglottic space. As she was busy, she could not advance treatment for a while. In March, 2010, she was referred to our department for further examination of a pharyngeal tumor that was in the tongue base.

Physical examination: Oral examination revealed a bulky, round and smooth mass in the lingual radix all around attached to the pharyngeal mucosa (Fig. 1A). A fiberscope showed a normal larynx and no recurrent nerve palsy was observed in the inferior portion of the epiglottis (Fig. 1B). Hematologic finding: FT3 2.3 pg/ml (2.1~3.8pg/ml), FT4 0.8 ng/dl (0.8~1.5ng/dl), TSH 11.36μU/ml (0.50~3.00μg/ml). Even though subclinical hypothyroidism was observed, Hashimoto's disease was not. The parathyroid hormone was normal. Thyroglobulin was high: 671 ng/ml (<32.7ng/ml).

Image finding: A computed tomography (CT) scan revealed a rounded mass with calcification in part in the foramen cecum occupying the pharyngeal space (Fig. 1C). No thyroid gland was seen in the normal pretracheal position (Fig. 1D). ^{99m}Tc sintigraphy showed high uptake in the sublingual region. On ²⁰¹Tl sintigraphy, wash out was poor which might have further complicated the malignant tumor (Fig. 1E).

Clinical diagnosis before surgery was ectopic lingual thyroid.

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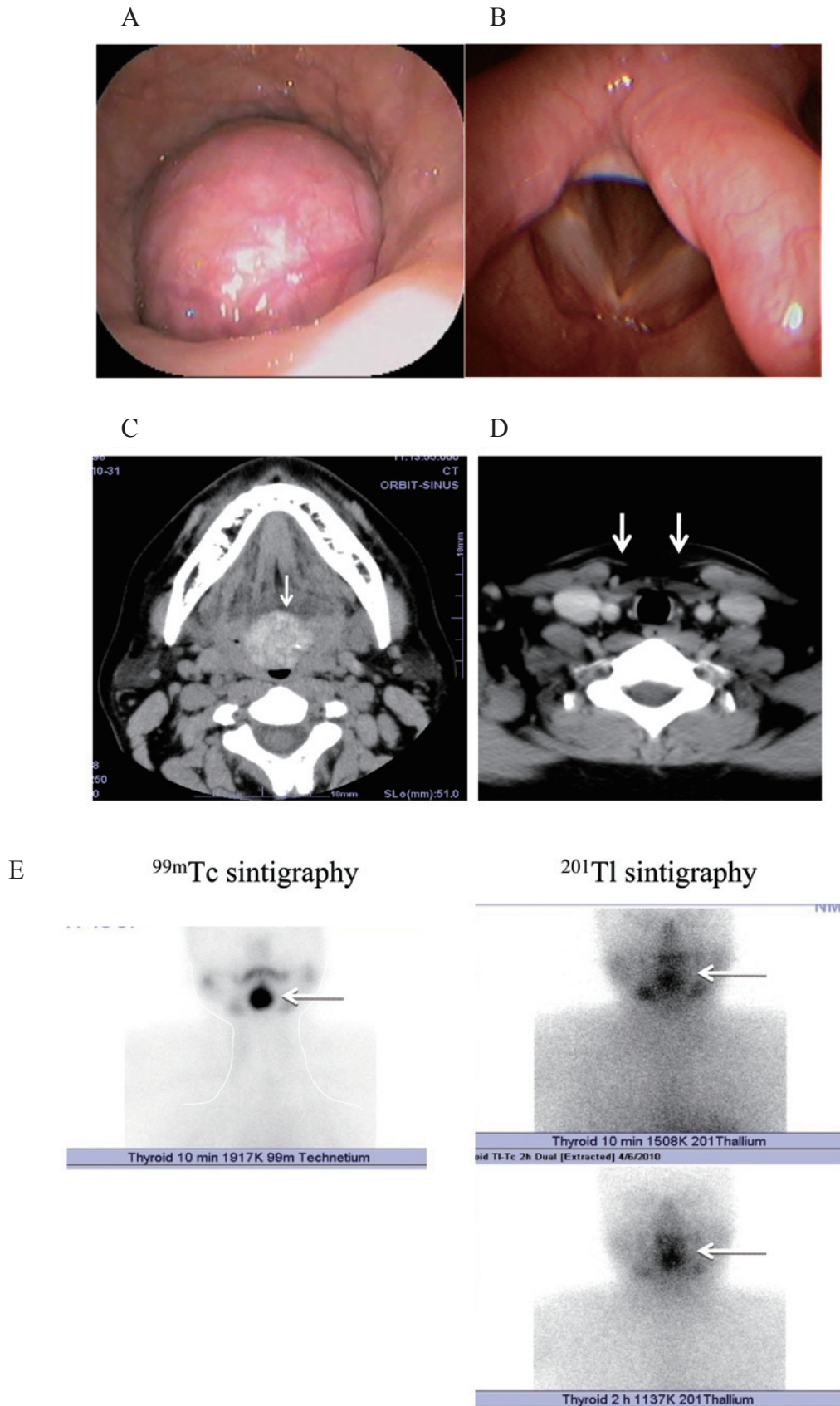


Fig. 1. Tumor formation observed in the oropharynx above the epiglottis (A). Fiberscope barely passed through the oropharynx and no obstruction was seen in the larynx (B). CT scan revealed that the tumor contains calcification (C). No structure was seen in the pretracheal region of the neck (D). ^{99m}Tc scintigraphy showed high uptake only in the sublingual region. In ^{201}Tl scintigraphy, wash out was poor at two hours after injection (E).

April 19, 2010, lingual thyroidectomy with an external cervical approach was performed under total anesthesia by conscious orotracheal intubation (Fig. 2A). A transverse incision was made in the neck above the hyoid bone (Fig. 2B). Suprahyoid muscles were exposed, and under the suprahyoid muscles, a round mass was seen. The lingual thyroid adhered to the pharyngeal mucosa and they were extracted together with thyroid tissue (Fig. 2C). After extraction, pharyngeal mucosa and mucosa were sutured in three layers. The tumor size was $30 \times 30 \times 25$ mm and was a round, solid mass (Fig. 2D). No accessory thyroid was observed. We also could not detect the feeding vessels which may have been one of the reasons for the dysfunctional thyroid gland.

The final histological examination confirmed that no malignancy was found in the mass and the

structure of the mass was the same as a normal thyroid (Fig. 2E).

Post surgery: steroid hormone was injected to avoid the formation of a pharynx edema. There was no apparent respiratory discomfort. There was a slight edema in the lingual radix but no air-way was obstructed (Fig. 3A). Intact PTH was 48 pg/ml (10~65pg/ml), which is a normal value. The thyroglobulin level was 12.3 ng/ml which subsided after surgery. The patient received nutrition and thyroid hormone from a feeding tube one day after surgery. 10 days after surgery, she started normal ingestion and was discharged on day 11 (Fig. 3B)

DISCUSSION

The ectopic thyroid is a congenital anomaly that is caused by undescendence of the embryonic thy-

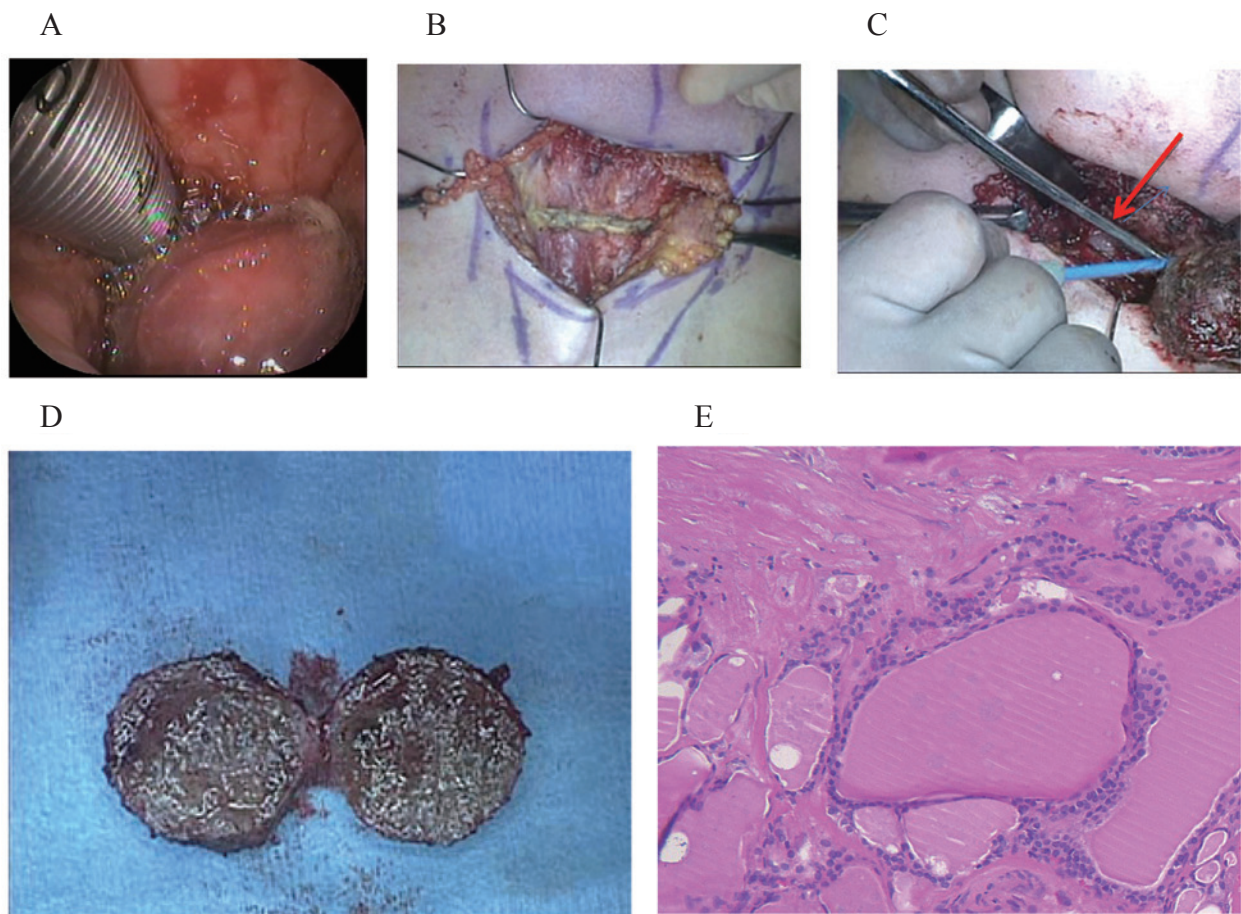


Fig. 2. Nasal intubation using a fiberscope was performed while the patient was conscious (A). The cervical approach was selected because of the size of the tumor. The hyoid bone was detected; the lingual thyroid was found above the hyoid bone (B). The lingual thyroid adhered to the pharyngeal mucosa and they were extracted together with thyroid tissue (C). The mass that was removed was round and solid (D). Histological examination revealed that no malignancy was found in the mass and the structure of the mass was the same as a normal thyroid (E).

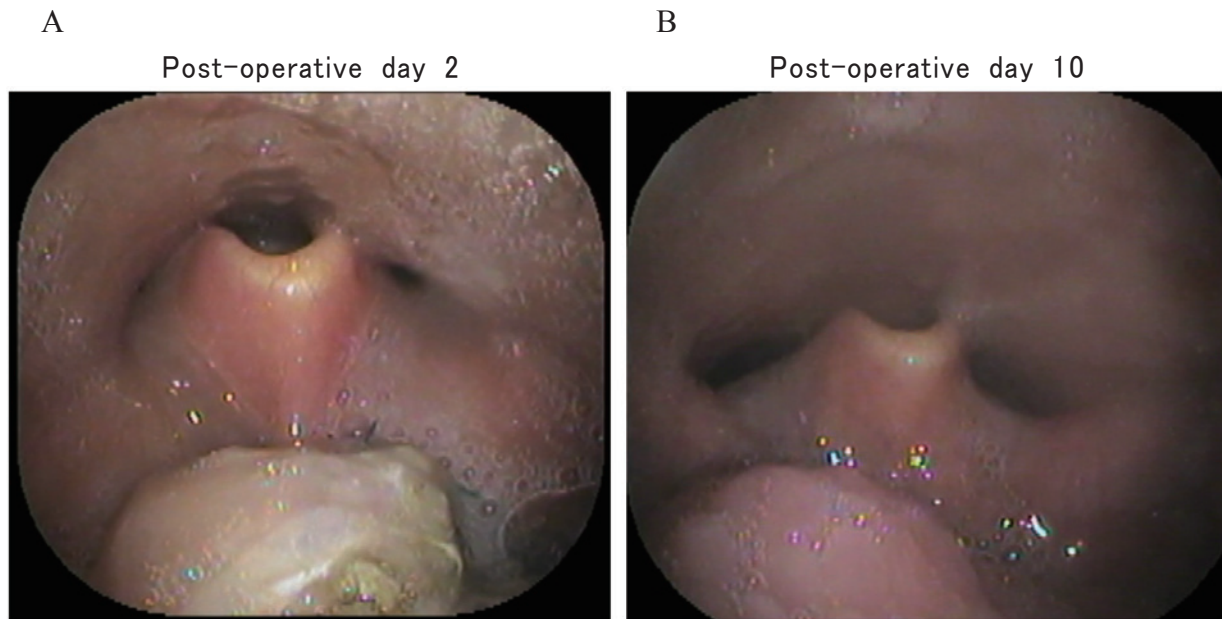


Fig. 3. The epiglottis was easily confirmed with a fiberscope after surgery. Slight edema and a white-coated suture was observed on day two after surgery but diminished by 10 day after surgery.

roid gland or aberration of the glandula thyroidea into the ductus thyroglossus [1][2]. A normal thyroid develops from the first and second branchial arch and most ectopic thyroids exist between the foramen cecum and the pretracheal position. However, some ectopic thyroids are found in the submandibular legion or lateral neck legion, thus a detailed outbreak mechanism is not totally understood. Some patients show a decrease in thyroid hormone while others show normal thyroidal function with potential hypothyroidism in which TSH decreases [4]. Ectopic thyroid is more likely to be found in females at puberty or during pregnancy [5]. The patient had become pregnant few years before she went to the hospital. This might be the reason why she had no symptom in her younger days. Thyroidal sintigraphy is the most specific examination for ectopic thyroid because the thyroid uptakes ^{99m}Tc and shows hot spots. One treatment for hypothyroidism involves taking a dosage of thyroid hormone which would reduce the volume of the tumor [6]. Hosokawa *et al.* reported that, following operation therapy, patients may show symptoms such as respiratory discomfort when a malignant tumor is suspected or when conservative treatment does not improve the symptoms [7]. There are reports of

ectopic thyroid carcinoma [5][8][9]. In our case report, the patient showed no respiratory discomfort. We opted for surgical therapy due to apprehension of occlusion of the pharynx and a possibility that the lingual thyroid could complicate the malignant tumor since the CT scan revealed calcification in the tumor, Tl sintigraphy revealed that wash-out was poor and the serum thyroglobulin level was high.

A tracheotomy might be required during an operation [10]. However, in our case, a fiberscope was nasally intubated while the patient was conscious. This is a recently improved anesthesia technology. In terms of management after the operation, although a steroid (betamethasone 2 mg/day) was injected, no edema was seen around the lingual portion. Furthermore, the patient did not complain about respiratory discomfort, and there were no complications such as a pharyngeal fistula. She was discharged 2 weeks after surgery. There are two ways to remove a tumor: a transoral approach and a cervical approach above the hyoid bone. The transoral approach is less invasive and leaves no scar on the neck [11] but the tumor is difficult to observe when it is large. Furthermore, in our case, because lingual thyroid was a suspected malignant

tumor, it had to be completely excised, hence the cervical approach. The tumor adhered to the pharyngeal mucosa and the adherent mucosa was extracted together with the ectopic thyroid. A stump of mucosa was sutured in 3 layers and the wound healed without making a pharyngeal fistula.

The accessory thyroid develops from the third and fourth branchial arch and is different from the thyroid [12]. In our case, the values of intact PTH were not different between the pre- and post-operative stage and hypocalcemia was not observed after the operation.

In conclusion, We experienced a case of ectopic lingual thyroid, which is large enough to be removed. Lingual tumor is rare but there is a possibility to cause airway constriction and ectopic lingual thyroid is one of the lingual tumor to be distinguished. We have performed surgery. The patient is required to take thyroid hormone every day for the rest of her life. However, she has no risk of suffocation anymore. Surgery is effective for the treatment of ectopic thyroid if possible.

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