

# A Case of Ectopic Lingual Thyroid with Situs Inversus Totalis

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Situs inversus totalis (SIT) is a rare congenital condition in which the viscera are transposed as a mirror of normal physiologic arrangement. We describe a rare case of lingual thyroid associated with SIT. A 64-year-old female who was diagnosed with SIT, visited our clinic due to pharyngeal foreign body sensation. Physical examination revealed a 2.5×2.5×2.0 cm sized, pinkish, round mass in the base of the tongue. Computed tomography suggested the diagnosis of lingual thyroid. She has been on regular follow up with levothyroxine therapy. To our best knowledge, this is the first case of lingual thyroid (LT) associated with SIT. The literature is reviewed and clinical features about LT and SIT are presented.

**Key Words:** Situs inversus, Lingual thyroid

## Introduction

Situs inversus totalis (SIT) is a rare congenital condition in which the viscera are transposed as a mirror of normal physiologic arrangement. The incidence is thought to be in the region of 1 : 5000 to 1 : 20,000 with a male/female ratio of 3 : 2.<sup>1)</sup> SIT is associated with various congenital anomalies. Lingual thyroid (LT) is a rare developmental thyroid anomaly usually affecting females. It is usually located in the midline and in the base of the tongue.<sup>2)</sup> The incidence of LT is reported as 1 : 100,000. In about 70% of the patients with LT there is an absence of normal thyroid gland.<sup>3)</sup> There are many papers reporting the association of SIT with congenital anomalies of various cardiovascular and other organ systems. But, there has been no report of SIT associated with LT. In this paper we present a case of LT with SIT who visited our clinic due to foreign body sensation on pharynx.

## Case Report

A 64-year-old female patient was admitted with complaint of pharyngeal foreign body sensation for several months. According to her medical history, she was diagnosed with situs inversus totalis. There was no history of thyroid problem, neck irradiation. Physical examination revealed a 2.5×2.5×2.0 cm sized, pinkish, round mass in the base of the tongue (Fig. 1). The thyroid gland was non-palpable and there was no palpable lymph node on neck. Chest X-ray and axial CT scan of the neck with contrast enhancement showed dextrocardia (Fig. 2). Neck CT scan showed 2.5×2.5×2.0 cm sized well circumscribed and well enhancing round mass in the posterior mid-portion of the tongue (Fig. 3), but showed no cervical thyroid gland in the usual pretracheal area (Fig. 4). According to the results of thyroid function tests, she was euthyroid state (T3: 1.49 ng/ml [Reference range, 0.8–2.0

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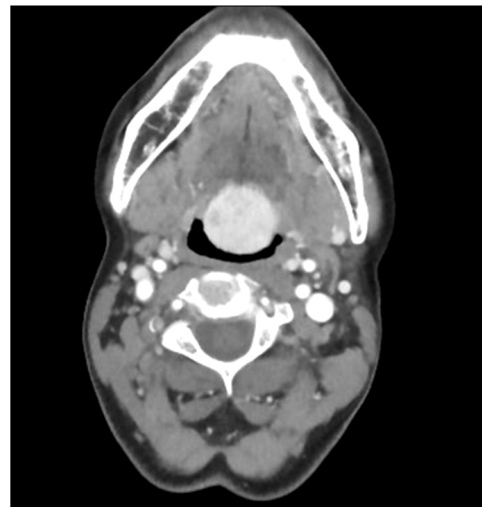
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ng/ml]; free T4: 12.8 pmol/L [Reference range, 11.5–23 pmol/L]; TSH: 1.21 mIU/L [Reference range, 0.17–4.05 mIU/L]). Other laboratory tests were within normal limits. Suppression treatment with levothyroxine was given and regular out-patient department follow up was planned.

## Discussion

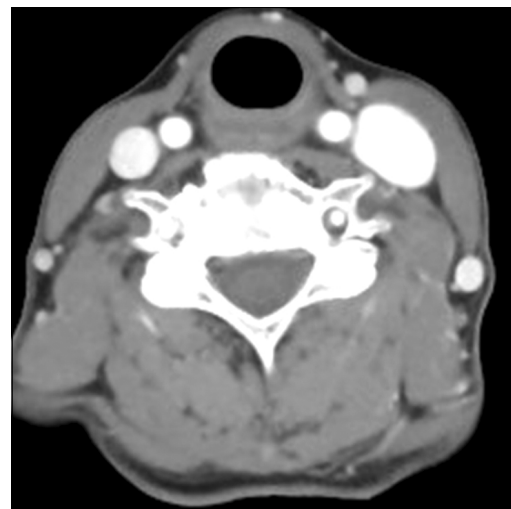
Dextrocardia is an abnormal congenital positioning of heart. Instead of heart forming in fetus on left side, it flips over and forms on right side. There are several types of dextrocardia. With situs solitus, one may have dextrocardia with normally related great arteries. With situs inversus (also known as SIT), one may have



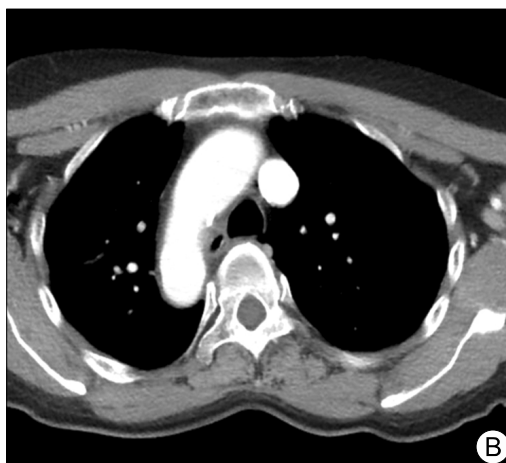
**Fig. 3.** Axial CT scan of the neck with contrast enhancement. It shows 2.5×2.5 cm sized well circumscribed and well enhancing round mass in the posterior midportion of the tongue.



**Fig. 1.** Laryngoscopic finding. It shows 2.5×2.5 cm sized pinkish round soft tissue mass in the base of tongue.



**Fig. 4.** Axial CT scan of the neck with contrast enhancement. It shows no cervical thyroid in the usual pretracheal area.



**Fig. 2.** Chest PA (A) and axial CT scan of the neck with contrast enhancement (B) show dextrocardia.

dextrocardia with inversely related great arteries. With situs ambiguous (either polysplenia or asplenia), one may have dextrocardia with any of the preceding relationships between the ventricles and great vessels.<sup>4)</sup> SIT is the most common type of dextrocardia in the general population. It is thought to occur in 1 to 2 per 10,000 population with equal frequency in males and females.<sup>5)</sup> The incidence of congenital heart disease is low, ranging from 2% to 5%.<sup>6)</sup> Approximately 60% of patients with SIT have other congenital abnormalities of the gastrointestinal tract including rotation anomalies, with or without volvulus, biliary atresia, splenic agenesis and colon aganglionosis.<sup>7)</sup>

Ectopic thyroid is an uncommon embryological aberration characterized by the presence of thyroid tissue in a site other than its usual pretracheal region. 90% of all ectopic thyroids are found to be lingual thyroids. Lingual thyroid is defined as the presence of thyroid tissue in the midline at the base of the tongue anywhere between the circumvallate papillae and the epiglottis.<sup>8)</sup> Up to 70% of patients with lingual thyroid have hypothyroidism.<sup>9)</sup> In our case, the patient was euthyroid state. Levothyroxine therapy corrects hypothyroidism and also induces shrinkage of gland.<sup>10)</sup> Surgical excision is an effective treatment for lingual thyroid, but no treatment should be attempted until I-131 radionuclide scan has determined that there is adequate thyroid tissue in the neck.<sup>10)</sup> In our case, she was put on levothyroxine therapy. She has been on regular follow up and has shown significant improvement symptomatically and some regression of the midline mass.

Although SIT is commonly associated with abnormalities of cardiovascular and other organ systems, the association between LT and SIT is not well understood. To our best knowledge, this is the first case of LT associated with SIT.

## Conclusion

Lingual thyroid is defined as the presence of thyroid tissue in the midline at the base of the tongue anywhere between the circumvallate papillae and the epiglottis. Although SIT is commonly associated with abnormalities of cardiovascular and other organ systems, the association between LT and SIT is not well understood. This is the first case of LT associated with SIT.

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