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Retrospective Case Series

## Prevalence of Dual Thyroid Ectopy in a Surgical Patient Population

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### Abstract

**Objectives:** Dual ectopic thyroids are considered rare events previously reported to occur in 9% of patients with ectopic thyroids. The objective of this study is to report an observed elevated prevalence of dual thyroid ectopy detected in patients presenting for surgical treatment of ectopic thyroids.

**Study Design:** Retrospective case series.

**Methods:** Surgical records at three tertiary academic referral centers were searched to identify patients undergoing ectopic thyroid excision from 1994 - 2012. Medical records and radiographic studies were reviewed by two independent reviewers.

**Results:** During the study period, 10 patients underwent surgical excision of ectopic thyroid tissue. Dual foci of ectopic thyroid tissue were identified in 4 patients. In all cases, one focus of ectopic tissue was lingual in location and the second focus was infrahyoid. No patients had an orthotopic gland.

**Conclusions:** A high prevalence of dual thyroid ectopy was detected in our patient population likely secondary to the use of comprehensive imaging studies in all patients. This suggests that a thorough evaluation of patients with ectopic thyroid tissue may identify additional occult foci.

**Keywords:** Ectopic Thyroid; Dual Ectopic Thyroid; Multifocal Ectopic Thyroid; Lingual Thyroid

### Abbreviations

CT: Computerized Tomography;

MRI: Magnetic Resonance Imaging;

FNA: Fine Needle Aspiration

## Introduction

Ectopic thyroids are rare developmental abnormalities created by aberrant descent of the thyroid from its origin at the base of the tongue. The estimated prevalence of thyroid ectopy is 1:100,000 - 300,000 [1]. Although ectopic thyroid tissue can occur throughout the body, most commonly it is located along the path of normal developmental descent including lingual (77 - 90%) and midline cervical locations (10 - 33%) [2,3]. Within this patient population, dual foci of ectopic tissue are infrequent and the literature is primarily limited to case reports and small series. The prevalence of dual thyroid ectopy has previously been estimated at 9% [4]. We present our 18 year experience with ectopic thyroids and elevated prevalence of dual ectopy.

## Materials and Methods

A retrospective chart review of patients from three tertiary academic referral centers undergoing surgical treatment for thyroid ectopy was conducted for the years 1994 - 2012. Patients were identified through a search of electronic surgical records for the terms: lingual thyroid, thyroid lingual, ectopic thyroid, thyroid ectopic, thyroid base of tongue, base of tongue thyroid, thyroid with transoral, and thyroid with laryngoscopy. Inclusion criteria mandated patients undergo surgical treatment of ectopic thyroid tissue with pathologic confirmation of diagnosis. Patients with ectopic thyroids undergoing medical management were excluded from the study due to a lack of systematic identification and the potential to create a biased or incomplete study population. The medical records and diagnostic imaging of all identified patients were analyzed by two independent reviewers for the presence of dual ectopic foci. The senior author reviewed and adjudicated any discrepancy between the two primary reviewers. This study was approved by the center's institutional review board.

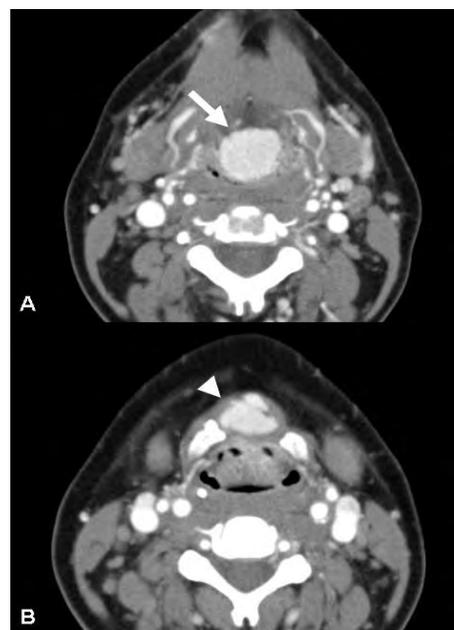
## Results

Ten patients were identified undergoing surgical treatment of ectopic thyroids during the years 1994 - 2012. The mean age was 37 years (range 11 - 59 years) with a 9:1 male to female ratio. All patients underwent preoperative imaging with CT, MRI, and/or Sestimibi. Dual foci of ectopic thyroid tissue were identified in 4 out of the 10 patients. One patient presented with an infrahyoid mass suspicious for follicular neoplasm on FNA and a symptomatic lingual thyroid; dual foci were confirmed on outside CT and Sestimibi scan. One patient presented with a symptomatic lingual thyroid and a preoperative CT scan identified an additional asymptomatic infrahyoid focus of ectopic thyroid (Figure 1). Two patients presented with symptomatic lingual thyroids and a history of prior infrahyoid ectopic thyroid excision. On preoperative imaging, one of the two patients had a persistent focus of ectopic tissue at the hyoid bone in addition to the lingual focus (Figure

2). No patients had orthotopic thyroid tissue. All patients underwent lingual thyroidectomy. A concurrent Sistrunk procedure was performed on the patient with suspicion for follicular neoplasm on FNA of their infrahyoid ectopic focus. All final pathology was consistent with benign thyroid tissue.



**Figure 1.** A 59 year old female noted by PCP to have anterior midline neck mass. FNA showed follicular thyroid pattern concerning for neoplasm. CT scan showed dual foci of thyroid ectopy (white arrows).



**Figure 2.** A) 48 year old female with a history of Sistrunk procedure presented with a lingual thyroid (white arrow) and B) persistent mid-

line cervical ectopic thyroid tissue (white arrowhead).

## Discussion

The majority of reports in the literature suggest that dual thyroid ectopy is a rare event estimated to occur in 9% of patients with ectopic thyroid tissue [4]. In comparison, we found dual foci of ectopic thyroid tissue in 40% of patients presenting for surgical treatment of thyroid ectopy. A recent report by Jain et al [5] similarly identified multifocal ectopic thyroid tissue in 5 out of 8 patients (62.5%).

Wildi-Runge et al's study represents the largest case series evaluating dual thyroid ectopy [4,5]. They evaluated 81 neonates with congenital hypothyroidism secondary to thyroid ectopy using pertechnetate scintigraphy and identified 7 cases (9%) with dual thyroid ectopy. This study's decreased prevalence of dual ectopy detection may be secondary to the lower sensitivity of 99mTc scintigraphy in the newborn period. In up to 76% of congenitally hypothyroid neonates without thyroid tissue on scintigraphy, thyroid tissue can in fact be identified on ultrasound exam [6]. Similarly, 92% of patients without thyroid tissue on scintigraphy in the newborn period will have identifiable uptake on repeat exam at 1 year of age [7]. Thus, Wildi-Runge et al's reported 9% prevalence most likely underestimates the true occurrence of dual thyroid ectopy.

A possible contribution to the recent elevated detection of dual thyroid ectopy is the increased use of comprehensive imaging. Historically, patients with ectopic thyroid tissue have not undergone extensive radiographic workup and small occult foci of ectopic tissue may have been overlooked. This is especially true in the pediatric population given the concerns of radiation exposure [8]. In our case series, all patients obtained a preoperative CT, MRI, and/or Sestimibi scan allowing for identification of asymptomatic foci. Likewise, all patients in Jain et al's study underwent Tc-99m Pertechnetate thyroid scans [5]. Based upon these results, the use of comprehensive imaging has improved our detection of dual thyroid ectopy and suggests that this condition occurs at a higher prevalence than previously believed.

Identification of dual thyroid ectopy is significant for patient counseling, although detection does not necessitate treatment. In our case series only one out of the three patients presenting with current dual ectopy underwent surgical treatment for both foci of thyroid tissue. However, all patients were counseled preoperatively that untreated foci of ectopic tissue has the potential to hypertrophy and become symptomatic or undergo neoplastic transformation, potentially requiring intervention in the future. This potential is evidenced in our own patients and case reports of ectopic thyroid tissue manifesting after total thyroidectomy [9,10]. Theoretically, circulating TSH recruits and stimulates the remaining ectopic thyroid tissue resulting in symptomatic hypertrophy [11]. Hence, awareness

and identification of dual ectopy during diagnostic evaluation improves a practitioner's ability to fully counsel and educate patients.

One weakness of this study is its small sample size and retrospective design. The increased prevalence of dual ectopic thyroids identified in this report may be secondary to sample bias from the small sample size. Thus all conclusions drawn from this study must be weighed against other reports in the literature.

## Conclusions

Dual thyroid ectopy may be historically underestimated and more prevalent than previously reported. The improved detection of dual ectopic thyroids in recent reports is likely secondary to the increased use of comprehensive imaging. Thus, a thorough evaluation of patients presenting with ectopic thyroid tissue may identify additional occult foci and improve patient counseling.

## References

1. Neinas FW, Gorman CA, Devine KD, Woolner LB. Lingual thyroid. Clinical characteristics of 15 cases. *Ann Intern Med.* 1973, 79(2): 205-210.
2. Kaplan M, Kauli R, Lubin E, Grunbaum M, Laron Z. Ectopic thyroid gland. A clinical study of 30 children and review. *J Pediatr.* 1978, 92(2): 205-209.
3. Larochelle D, Arcand P, Belzile M, Gagnon NB. Ectopic thyroid tissue – a review of the literature. *J Otolaryngol.* 1979, 8(6): 523-530.
4. Wildi-Runge S, Stoppa-Vaucher S, Lambert R, Turpin S, Van Vliet G et al. A high prevalence of dual thyroid ectopy in congenital hypothyroidism: evidence for insufficient signaling gradients during embryonic thyroid migration or for the polyclonal nature of the thyroid gland? *J Clin Endocrinol Metab.* 2012, 97(6): E978-E981.
5. Anuj Jain, Sujata Pathak. Rare developmental abnormalities of thyroid gland, especially multiple ectopia: A review and our experience. *Indian J Nucl Med.* 2010, 25(4): 143-146.
6. Deladoey J, Ruel J, Giguere Y, Van Vliet G. Is the incidence of congenital hypothyroidism really increasing? A 20-year retrospective population-based study in Quebec. *J Clin Endocrinol Metab.* 2011, 96(8): 2422-2429.
7. Chang YW, Lee DH, Hong YH, Hong HS, Choi DL et al. Congenital hypothyroidism: analysis of discordant US and scintigraphic findings. *Radiology.* 2011, 258(3): 872-879.

8. Ruth A. Kleinerman. Cancer risks following diagnostic and therapeutic radiation exposure in children. *Pediatr Radiol.* 2006, 36(2): 121-125.
9. Winters R, Christian RC, Sofferman R. Thyrotoxicosis due to ectopic lateral thyroid tissue presenting 5 years after total thyroidectomy. *Endocr Pract.* 2011, 17(1): 70-73.
10. Byrd MC, Thompson LD, Wieneke JA. Intratracheal ectopic thyroid tissue: a case report and review of the literature. *Ear Nose Throat J.* 2003, 82(7): 514-518.
11. Dossing H, Jorgensen KE, Oster-Jorgensen E, Krogdahl A, Hegedus L. Recurrent pregnancy-related upper airway obstruction caused by intratracheal ectopic thyroid tissue. *Thyroid.* 1999, 9(9): 955-958.