DUAL ECTOPIC THYROID: A CASE REPORT

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ABSTRACT
Dual ectopic thyroid is very rare. We are reporting a case of dual ectopic thyroid in the lingual and infrahyoid areas in a 16 year old female patient with no thyroid gland in its normal anatomical location. On physical examination, there was a 9 x 6 cm anterior midline neck swelling just below the hyoid bone and a 1.5 x 1 cm mass at the base of the tongue. Triiodothyronine (T₃) and thyroxine (T₄) levels were normal and thyroid-stimulating hormone (TSH) was marginally raised. A thyroid scan with technetium-99m sodium pertechnate confirmed dual ectopic thyroid with no iodine uptake in the normal anatomical location of the thyroid gland. The infrahyoid ectopic thyroid was surgically removed for cosmetic reasons, and the lingual thyroid, which was asymptomatic left untouched. The importance of thyroid scanning in the evaluation of anterior midline neck swellings and treatment options are discussed.

Key Words: Ectopic Thyroid, Lingual Thyroid

INTRODUCTION
Ectopic thyroid refer to all cases in which the thyroid gland is found in a location other than its usual site anterior to trachea in the lower neck (Baik et al., 2002). Ectopic thyroid tissue can be found anywhere from foramen cecum to all along the normal descent path upto the normal cervical position of the thyroid gland. The most common site of the ectopic thyroid is at the base of tongue followed by sublingual and the anterior midline of neck (Damiano et al., 1996). We are reporting a case with dual ectopic thyroid having lingual thyroid with infrahyoid ectopic thyroid.

CASES
A 16 year old female patient presented with history of swelling in the midline neck for 13 years (figure 1). The midline neck swelling initially was high in the neck (figure 2). The neck swelling has been gradually increasing in size for last 3 years with marked increase in size in last 1 year. No other associated symptoms like change in voice or difficulty in swallowing were complained except the cosmetic appearance. Her past and family histories were not significant. A presumptive diagnosis of thyroglossal cyst was made and the patient was admitted.

On examination there was a midline neck swelling extending from hyoid to supra sternal notch with no distortion of overlying skin. The swelling was soft, cystic, non-tender measuring 9 cms x 6 cms. It was freely mobile in all directions. Transillumination test was positive. There was no associated cervical lymphadenopathy. On indirect laryngoscopy small pinkish mass was observed at the base of tongue. On bimanual palpation firm, irregular surfaced, non-tender, 1.0 cm x 1.5 cm mass felt at the base of tongue. The remaining findings on physical examination were not remarkable. The T3 level was 1.01 ng/ml (0.60-1.81 ng/ml), T4 level was 6.3 µg/dl (4.5-10.9µg/dl) and the thyroid stimulating hormone level was 8.61µIU/ml (0.35-5.5µIU/ml). The raised TSH showed marginal asymptomatic hypothyroidism. Rests of the investigations were within normal limits. A contrast enhanced computarised tomography scan of the neck revealed absence of normal thyroid glandular tissue in thyroid bed. It showed a large well circumscribed lobulated cystic lesion having thin internal septation measuring 98 mm x 63mm x 52 mm extending from infra hyoid region to supra clavicular region. A markedly enhancing soft tissue lesion of
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Figure 1: Patient presented with large midline infrahyoid cystic swelling

Figure 2: Infra hyoid swelling at the age of 3 year

Figure 3: CECT neck coronal cut showing lingual thyroid and cystic mass at thyroid bed

Figure 4: Thyroid scan showing lingual thyroid and scattered thyroid tissue in cystic thyroid swelling

Figure 5: Excised cystic swelling
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size 18mm x 20 mm was also seen at the base of tongue extending towards vallecula (figure 3). A thyroid scan with Tc-99m sodium pertechnetate (figure 4) showed uptake in the region of base of tongue and patchy radiotracer uptake with multiple ill defined cold areas in midline neck swelling at thyroid bed extending upto suprasternal notch. Normal thyroid gland was not seen. Colloid was aspirated on Fine needle aspiration but on cytology follicular epithelial cells were not seen. Midline collar incision made. Superior and inferior subplatysmal flap rose. Strap muscle separated in midline. The cystic lesion in midline was extending from hyoid to suprasternal region. It was carefully dissected from all around. Normal thyroid gland was not seen in the thyroid bed. Cyst was excised and wound is closed in three layers after placing a corrugated rubber drain. The dimensions of cystic swelling were 10cms x 7cms x 5cms (figure 5). On histopathology cyst was filled with colloid, cyst wall showed scattered thyroid follicular epithelial cells. Post operative period was uneventful. Post operative thyroid function showed hypothyroidism so patient was kept on 100µgm levothyroxine. With this 3 year follow up showed no change in size of lingual thyroid with normal thyroid function test.

DISCUSSION

Thyroid primordium develops as an endodermal bud from the anterior floor of pharynx and descents down on either side of trachea where it fuses with the caudal prolongation of the forth pharyngeal pouch to form the thyroid gland. Arrest can occur at any site in this tract of descent leading to an ectopic thyroid (Hazarika et al., 1998). It is very unusual for two ectopic foci of thyroid tissue to present simultaneously. Congenital abnormalities are the least common of the thyroid disorders. An ectopic thyroid gland is reported to occur in 1 in 4000 to 8000 patients with thyroid disease (Baik et al., 2002). The male-female ratio is approximately 1:4 to 1:7 (Kansal et al., 1987). A simultaneous ectopia of thyroid gland at tongue base and cervical region is extremely rare.

Most of the ectopic thyroids are asymptomatic however may present with foreign body sensation, dysphagia or lump in throat (Hazarika et al., 1998). It has two modes of presentation based on patient age at presentation (Williams et al., 1996). One group is that of infants and young children who suffer from failure to thrive and mental retardation. The second consists of those at peripubertal ages and lingual thyroid presents in them with symptoms of oropharyngeal obstruction. This occurs because of hypertrophy of thyroid tissue due to increase in TSH level as a result of enhanced metabolic stress during puberty (Wertz, 1976) as was seen in our case.

About 10% of these patients present with hypothyroidism in spite of hyperplasia of gland (Baik et al., 2002). When both lingual and cervical ectopic thyroid coexist, the patient may often be euthyroid despite enlargement of the ectopic thyroid tissue. In our case, the patient presented with an elevated TSH level (8.61µIU/mL) with normal T3 and T4 levels. In most of these cases thyroid tissue is not found in its normal position. So surgical excision of this tissue, under the impression of a thyroglossal duct cyst will result in permanent hypothyroidism (Al-Dousary, 1997). Therefore a thyroid scan along with entire neck CT or neck USG should be performed routinely to avoid such a catastrophe (Morgan et al., 1995). Thyroid scintigraphy is highly specific and sensitive in detecting normal and ectopic thyroid (Sood et al., 2008). The present case was also diagnosed initially thyroglossal duct cyst at the first visit. The presence of ectopic thyroid could only be confirmed after further imaging studies.

Management of ectopic thyroid tissue is dependent on several factors including size of lesion, presence of local symptoms, age and status of thyroid gland and presence of complication factors like ulceration, hemorrhage or malignancy (Wertz, 1976, Baik et al., 2002). Options include hormone therapy, radioiodine ablations and surgery. Surgical excision is the mainstay of treatment of symptomatic ectopic thyroid. It should be taken care that surgery may however result in permanent hypothyroidism if the ectopic tissue is the only functional thyroid tissue in body. In our case, though the cervical ectopic was removed because of cosmetic reasons and cystic degeneration. The functional thyroid tissue in lingual thyroid was not sufficient enough to maintain euthyroid status of the patient. Patient is euthyroid on 100 µgm levothyroxine once a day with no enlargement of lingual thyroid in three years follows up.
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Conclusion
Ectopic thyroid is not so uncommon lesion, pre operative thyroid scan and postoperative thyroid function test is necessary to detect the functioning thyroid tissue.

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REFERENCES