Sublingual Ectopic Thyroid Gland Simulating A Thyroglossal Duct Cyst- A Case Report

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Ectopic thyroid is a rare embryological aberration of the thyroid descend in children. We present a case of ectopic thyroid simulating a thyroglossal cyst in a two year old boy, who was diagnosed intra-operatively. The patient was treated with L-thyroxine. We review the embryological features and the approach to the diagnosis and management of this congenital anomaly.

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Ectopic thyroid is a very rare developmental anomaly of descent of the thyroid gland¹. Most of the patients present with a asymptomatic midline neck mass and the diagnosis is easily mistaken for a thyroglossal cyst². Differentiation of these two conditions is extremely important, since removal of the sole functioning thyroid tissues result in permanent hypothyroidism. Therefore, many diagnostic tests including thyroid function test, ultrasound of the neck and thyroid scanning had been recommended in the preoperative evaluation of a thyroglossal cyst³. The necessity of these tests was questioned by many authors and still is a subject of controversy^{4,5}. Treatment options of ectopic thyroid include both hormonal therapy and surgical intervention, but the role of each option is not clearly identified⁶.

We present a child with a neck swelling which was thought initially to be a thyroglossal cyst, but intraoperatively found to be an ectopic thyroid gland.

THE CASE

A two year old boy presented to pediatric surgery clinic in September 2002 with a history of painless, midline neck swelling of three months duration. The swelling increased slightly in size, but there were no pressure symptoms. On clinical examination the patient was euthyroid, with normal growth and development. 1.5x1.5 cm smooth mobile well-circumscribed mass was palpable in the midline of the neck just below the hyoid bone. The thyroid gland was not palpable and no other cervical mass was found. Clinical examination of the other systems was unremarkable. This swelling was initially diagnosed as a thyroglossal cyst and the patient underwent surgery for excision. Intraoperatively, the mass was found to be solid and highly

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vascular. The pretracheal area was explored and no thyroid tissue was found in the normal anatomical location. Therefore, suspicion of ectopic thyroid was raised, the mass was left undisturbed and biopsy was taken, which revealed a normal thyroid tissue. Postoperatively, he was clinically euthyroid and thyroid function test showed increased thyroid stimulating hormone—and normal free thyroxine-4. After a one month therapy with L-thyroxine, thyroid function test was normalized and the swelling had significantly reduced in size. Thyroid scan was performed thereafter looking for other functioning thyroid tissue, which showed only a single midline sublingual ectopic thyroid tissue (Fig 1,2).

Figure 1. Thyroid scan shows no functioning thyroid tissue in the normal location, but an area of focal accumulation of the tracer is seen in the midline sublingual region (arrow), suggestive of ectopic thyroid gland.

DISCUSSION

Ectopic thyroid is an uncommon embryological aberration of the thyroid descend and characterized by the presence of thyroid tissue in a site other than its usual pretracheal region. It occurs along the path of descent of the developing thyroid primordium from the foramen cecum. Most commonly, the gland completely fails to descend and presents as a *lingual thyroid* and this is the only thyroid tissue in 70% of cases¹. This is in contrast to a *sublingual thyroid*; in which case, there is incomplete descent of the gland where the final resting point may be high in the neck or just below the hyoid bone⁷. When the thyroid gland is located in its normal position in the lower neck, fragments of the thyroid tissue representing *ectopic thyroid* may still be found anywhere along its course.

More than 400 cases of lingual thyroid have been documented in the literature, but the reported cases of sublingual thyroid is substantially lower^{7,8}. Ectopic thyroid is subject to same diseases as the anatomically correctly positioned thyroid, such as nodular hyperplasia and rarely neoplastic degeneration⁹. Asymptomatic mass lesion is the usual presentation of ectopic thyroid, but obstructive symptoms, hypothyroidism and very rarely hyperthyroidism have been documented².

The inadvertent removal of ectopic thyroid gland that was mistaken for thyroglossal duct cyst leading to significant hypothyroidism has been reported in the literature¹⁰. Therefore, differentiation of these two conditions is extremely important. To prevent this problem, routine preoperative identification of the normal thyroid gland by an ultrasound is advocated in all cases of thyroglossal cyst⁴. When the thyroid gland can be identified in the normal position, coexistent ectopic thyroid is seldom found⁵. Thyroid scintigraphy is the best method in identifying all sites of functioning thyroid tissue, but routine thyroid scan is not necessary¹¹. It is justified in cases of ectopic

thyroid and where normally located thyroid gland cannot be detected³. Thyroid function test is an essential test and should be included in the evaluation of all cases of ectopic thyroid⁵.

Many authors agree that hormonal suppressive therapy and preservation of the ectopic tissues are the primary goals in the treatment⁶. Surgery is reserved for those cases of failure of medical therapy in symptomatic patient or in cases of rare malignant degeneration⁶.

The patient presented with a asymptomatic mass mimicking thyroglossal cyst and the preoperative diagnosis of thyroid ectopia was not suspected. Thyroid function test was done postoperatively and the result was consistent with compensated hypothyroidism. He would have avoided unnecessary surgery if a routine preoperative ultrasound study looking for the normal thyroid gland had been done.

CONCLUSION

Ectopic thyroid, though rare, should be one of the differential diagnoses in the midline neck masses in children. A preoperative ultrasound is helpful in distinguishing thyroglossal cyst from this congenital anomaly and prevents inadvertent removal of what may be the only thyroid tissue in these patients. We presented a case of sublingual ectopic thyroid simulating a thyroglossal cyst, which was diagnosed intraoperatively and treated with hormonal therapy.

REFERENCES

- 1. Baik SH, Choi JH, Lee HM. Dual ectopic thyroid. Eur Arch Otorhinolaryngol 2002:259:105-7.
- 2. Abdallah-Matta MP, Dubarry PH, Pessey JJ, et al. Lingual thyroid and hyperthyroidism: a new case and review of the literature. J Endocrinol Invest 2002;25:264-7.
- 3. Kessler A, Eviatar E, Lapinsky J, et al. Thyroglossal duct cyst: is thyroid Scanning necessary in the preoperative evaluation? Isr Med Assoc J 2001;3:409-10
- 4. Gupta P, Maddalozzo J. Preoperative sonography in presumed thyroglossal duct
 - cysts. Arch Otolaryngol Head Neck Surg 2001;127:200-2.
 - 5. Al-Dousary S. Current management of thyroglossal-duct remnant. J R Soc Med 2000;93:18-21.
- 6. Haddad A, Frenkiel S, Costom B, et al. Management of the undescended thyroid.
 - J Otolaryngol 1986;15:373-6.
 - 7. Hazarika P, Siddiqui SA, Pujary K, et al. Dual ectopic thyroid: a report of two cases. J Laryngol Otol 1998;112:393-5.
- 8. Quarracino MC, Aguas SC. Lingual Thyroid: A clinical case. Med Oral 2003;8:57-60.
 - 9. Pang YT. Ectopic Multinodular Goitre. Singapore Med J 1998;39:169-70.
- 10. Leung AK, Wong AL, Robson WL. Ectopic thyroid gland simulating a thyroglossal duct cyst: a cae report. Can J Surg 1995;38:87-9.

11. Lim-Dunham JE, Feinstein KA, Yousefzadeh DK, et al. Sonographic demonstration of a normal thyroid gland excludes ectopic thyroid in patients with thyroglossal duct cyst. Am J Roentgenol 1995;164:1489-91.