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Case Report

Ectopic thyroid tissue presenting as an external auditory canal mass

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ABSTRACT

An ectopic thyroid is a rare occurrence with a majority of ectopic thyroid tissue located in the lingual region or in the midline. The abnormal sites represent developmental defects in migration from the floor of the primitive foregut at the foramen caecum of the tongue to the final pre-tracheal position of the gland. A few cases of ectopic thyroid tissue have also been reported from sites seemingly unrelated to the normal development of the thyroid gland. We report a case of ectopic thyroid in the external ear canal, which presented as a small reddish mass in the external ear canal. To the best of our knowledge, this is the first such reported case in literature and adds to the body of knowledge in such cases. Although such a finding is exceedingly rare, the authors recommend routine histopathology in all cases of polyps in the external canal and standard investigation for the status of the residual thyroid gland via isotope scans, ultrasonography or thyroid function tests.

Keywords: Ectopic thyroid, External auditory canal

INTRODUCTION

An ectopic thyroid refers to the presence of thyroid tissue in locations other than the normal anterior neck region. Abnormal location of thyroid tissue occurs as a result of aberrant migration from the formaen caecum to the level of the 2nd or 3rd tracheal ring, which is the final position of the thyroid gland, with a general prevalence of 1 in 100000 to 1 in 300000.[1,2] Lingual thyroid is the most frequent ectopic location of the thyroid gland accounting for 90% of cases, though ectopic thyroids have also been reported from the submandibular region mediastinum and heart trachea and intrathoracic region.[3-11] Reported abdominal locations include the gall bladder and the adrenal gland.[12,13] To date, there has been no case report of ectopic thyroid tissue in the ear canal. We report a case of normal thyroid tissue presenting as a mass in the external ear canal and offer possible reasons for this rare occurrence.

CASE REPORT

A seventeen year old female, with no known co-morbidities or allergies, presented to the Otorhinolaryngology clinic of a busy tertiary hospital (Hospital Melaka) with left ear pain for the past one month. There were no other ear symptoms. She had sought treatment from a general practitioner who prescribed antibiotic eardrops which did not alleviate symptoms. She had no nasal or throat symptoms. Her menstrual history was normal, sleep and appetite were not disturbed and bowel and bladder habits were normal.

On otoscopic examination of her left ear, we noted a single reddish mass measuring about 6 mm × 6 mm on inspection located on the anterior wall of the lateral third of her external ear canal. There was no discharge and there was no mastoid or tragal tenderness. Tuning fork tests were normal. There were no abnormalities detected on right ear examination. Her nasal and throat examination was normal as well.

As per protocol, we proceeded with an excision biopsy under the microscope in the clinic and the excised tissue was inserted in formalin, labelled and a request for histopathology examination was obtained. There was minimal bleeding which was managed with a small temporary ear wick. The patient was discharged and called for follow-up after two weeks.

The histopathology report was as below:

Section shows a piece of keratinaceous cellular debris and a piece of thyroid tissue. The latter consists of a few isolated thyroid acini lined by benign follicular cells with some containing colloid. No atypia, granuloma, inflammation or malignancy seen. Features are in keeping with ectopic thyroid with branchial pouch/cleft anomalies.

At follow-up one-month post polypectomy, the patient was well with no recurrence and no residual symptoms. We proceeded with a baseline thyroid function test which was normal and an ultrasound of her thyroid gland revealed a normally situated thyroid gland. At six months, the patient is symptom-free and symptomatically and biochemically euthyroid. The photomicrographs of the slide is in Figure 1.

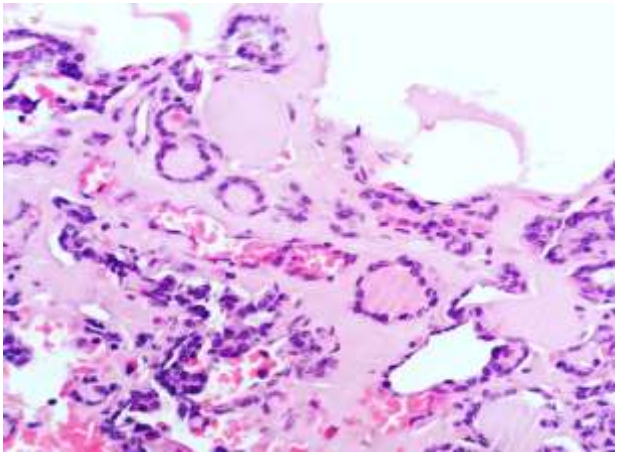

Figure 1: H-E stain of excised ear canal tissue showing thyroid acini.

DISCUSSION

Ectopic thyroid was first described by Hickman in 1869 in a newborn who suffocated sixteen hours after birth because of a lingual thyroid causing upper airway obstruction.[14] Although the general reported prevalence is in the range of 1 in 100000 to 1 in 300000, autopsy studies indicate the prevalence may be much higher, in the range of 7-10%.[5,15] Around 500 cases have been reported so far with the commonest site is lingual accounting for 90% of cases.[16]

The human external ear canal develops from the pharyngeal arch system which is involved in various aspects of the development of the head and neck. The arches are separated from each other by a groove, or cleft, and the primitive pharynx develops outpouchings called pouches. Though there is no communication between the pharynx and the clefts, the clefts come into close contact with the pouches.[17]

The primitive external auditory canal is formed by invagination of the upper or dorsal portion of the first pharyngeal cleft canal. At 4 to 5 weeks' gestation, which is around the time the primitive thyroid gland is formed and starts its descent, the primitive external canal establishes contact with the first pharyngeal pouch. This contact is soon lost due to proliferation of embryonic connective tissue, which separates the two structures.[17]

The thyroid gland is located in the anterior neck region between the second and fifth tracheal rings. It is the first of the bodies endocrine glands to develop on approximately the 24th day of gestation. There are two types of cells in the mature thyroid gland. The follicular cells are derived from the thyroid anlage which originates as a proliferation of endodermal epithelial cells on the median surface of the developing pharyngeal gut between the first and second pharyngeal pouches.[17] The C-cells originate from the ultimo-branchial bodies of the fourth pharyngeal pouch. The thyroid follicular cells are responsible for thyroid hormone production, while the C-cells produce calcitonin.[18]

In the third or fourth week of gestation, an endodermal diverticulum from the floor of the pharyngeal gut is formed. This diverticulum descends in the midline, from the foramen cecum to the final location of the gland. This migration begins at embryonic day 24, just before the formation of the external auditory canal resulting in the formation of the thyroglossal duct. Ectopic thyroid tissue is the result of a failure of migration of thyroid which is usually along the route of thyroglossal duct.

The mechanisms responsible for thyroid morphogenesis were described by De Felice and Lauro.[18] In particular, transcription factor TITF1/NKX2-1, which is responsible for the thyroid specific expression of thyroglobulin (Tg) and thyroperoxidase and the transcription factors PAX8, HHEX, and FOXE1 are implicated in being essential for the early stages of thyroid morphogenesis. TITF1/NKX2-1 controls survival at the beginning of organogenesis as well as the expression of genes specific for thyroid follicular cells in adult life.[18] PAX8 is required for the survival of thyroid cell precursors and for their functional differentiation. It plays a key role in the genetic regulatory cascade, which controls thyroid development.[18,19] It is of note that some cases of thyroid dysgenesis may be due to mutations in genes regulated by the aforementioned transcription factors.[17] HHEX may be required to maintain the expression of TITF1/NKX2-1, FOXE1, and PAX8 mRNA in the thyroid anlage. Genetic studies have demonstrated that FOXE1 it is required for thyroid migration in mice, but no mutations

have been detected in humans till date.[17] Based on the above genetic factors and the hypothesis that that a mutation may have occurred in one of the transcription and regulatory proteins listed, we speculate that the abnormal location of thyroid tissue in our patient was the result of faulty migration or implantation in areas anatomically close during early embryogenesis.

CONCLUSION

Ectopic thyroid tissue is a rare entity. The lesion may present in areas away from the midline or in the line of the descending thyroglossal duct and requires an awareness of reported sites and presentations to be diagnosed. All cases of ectopic thyroid tissue must be investigated for the location and function of normal thyroid tissue as its inadvertent removal may render the patient hypothyroid. In our case, due to a low index of suspicion and the small size of the mass, this investigation was done after tissue diagnosis of the excised mass. The presence of tissue in the external ear canal was surprising and is rare but we speculate that such lesions are underdiagnosed because of a low index of suspicion and inconsistent protocols to send tissues for histopathology.

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REFERENCES

1. Di Benedetto V. Ectopic thyroid gland in the submandibular region simulating a thyroglossal duct cyst: a case report. J Pediatric Surg. 1997;32:1745-6.
2. Babazade F, Mortazavi H, Jalalian H, Shahvali E. Thyroid tissue as a submandibular mass: a case report. J Oral Sci. 2009;51:655-7
3. Toso A, Colombani F, Averono G, Aluffi P, Pia F. Acta Otorhinolaryngol Ital. 2009;29(4):213-7.
4. Kumar R, Sharma S, Marwah A, Moorthy D, Dhanwal D, Malhotra A. Ectopic goiter masquerading as submandibular gland swelling: a case report and review of the literature. Clin Nucl Med. 2001;26:306-9.
5. Bersaneti JA, Silva RD, Ramos RR, Matsushita Mde M, Souto LR. Ectopic thyroid presenting as a submandibular mass. Head Neck Pathol. 2011;5:63-6.
6. Pollice L, Caruso G. Struma cordis. Ectopic thyroid goiter in the right ventricle. Arch Pathol Lab Med. 1986;110:452-3.
7. Porqueddu M, Antona C, Polvani G, Cavoretto D, Pompilio G, Gianolli L, et al. Ectopic thyroid tissue in the ventricular outflow tract: embryologic implications. Cardiology. 1995;86:524-6.
8. Ferlito A, Giarelli L, Silvestri F. Intratracheal thyroid. J Laryngol Otol. 1988;102:95-6.
9. Muysoms F, Boedts M, Claeys D. Intratracheal ectopic thyroid tissue mass. Chest. 1997;112:1684-5.
10. Yang Y, Li Q, Qu J, Xiang Y, Pan Y, Liao Z, et al. Ectopic intratracheal thyroid. Southern Med J. 2010;103(5):467-70
11. Sakorafas GH, Vlachos A, Tolumis G, Kassaras GA, Anagnostopoulos GK & Gorgogiannis D. Ectopic intrathoracic thyroid: case report. Mount Sinai J Med. 2004;71(2):131-3.
12. Cassol CA, Noria D, Asa SL. Ectopic thyroid tissue within the gall bladder: case report and brief review of the literature. Endocrine Pathol. 2010;21:263-5.
13. Shuno Y, Kobayashi T, Morita K, Shimizu S, Nishio Y, Ito A, et al. Ectopic thyroid in the adrenal gland presenting as cystic lesion. Surgery. 2006;139(4):580-2.
14. Bianco MR, Boria AL, Franco T, Ferrise P, Eugenia Allegra. Ectopic lingual thyroid with vascular anomalies. Int Med Case Rep J. 2013;6:55–8.
15. Kousta E, Konstantinidis K, Michalakis C, Vorias M, Sambalis G, Georgiou M, et al. Ectopic thyroid tissue in the lower neck with a coexisting normally located multinodular goiter and brief literature review. Hormones. 2005;4:231-4.
16. Basaria S, Westra WH, Cooper DS. Ectopic lingual thyroid masquerading as thyroid cancer metastases. J Clin Endocrinol Metabolism. 2001;86:392-5.
17. Wright CG. Development of the Human External Ear. J Am Acad Audiol. 1997;8:379-82.
18. De Felice M, Lauro R. Thyroid development and its disorders: genetics and molecular mechanisms. Endocrine Rev. 2004;25:722-46.
19. Pasca di Magliano M, Di Lauro R, Zannini MS. PAX8 has a key role in thyroid cell differentiation. PNAS. 2000;97(24):13144-9.

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