A rare presentation of ectopic thyroid gland at right axilla

Yik Hin Chin, MRCP, Yon Lek Yap, MRCP, Huang Hin Chin, MRCP, Sarina Salim, M. Med Radiology

1 Hospital Sultanah Nora Ismail, Batu Pahat, Johor Malaysia, 2 Hospital Enche Besar Hajjah Kalsom, Kluang, Johor, Malaysia

SUMMARY
The thyroid gland and its hormones play important roles in organ development and in the homeostatic control of physiological mechanisms in human beings. As a result of embryogenic descent of thyroid gland, it commonly resides along the midline – from tongue to mediastinum (90%). Ectopic thyroid gland is a rare occurrence, with extra-lingual ectopic thyroid gland being even rarer. Thus, there is a concern for malignant metastasis. Madam H, a 56-year-old healthy woman presented to the Hospital Sultanah Nora Ismail, Johor, Malaysia in April 2020 with an increasing size of right axilla mass and history of weight loss. She was having right axilla mass for the previous 7 years but only noticed the increase in size about 1 year ago. She has no other constitutional symptoms. A tru-cut biopsy performed demonstrated a benign ectopic thyroid tissue. Thyroid function test showed primary hypothyroidism. Serum Chromogranin A and other thyroid antibodies were within the normal value. Further radiological imaging showed the normal thyroid gland at neck, with no signs of distant malignancy. There was no other axillary, mediastinal or hilar lymph node enlargement. She was started on regular T. L-Thyroxine 100mcg OD (maintenance) in May 2020. The titration of L-thyroxine was done subsequently in visiting endocrine clinic. Benign ectopic thyroid gland is an unusual finding. As such, follow up is needed with possibility of carcinomatous transformation such as papillary carcinoma should be considered.

INTRODUCTION
The thyroid gland and its hormones play multifaceted roles in organ development and in the homeostatic control of physiological mechanisms in human beings. During embryogenesis, thyroid gland descends from the floor of the primitive foregut to the final position of the thyroid in the neck. As a result, ectopic thyroid gland can reside anywhere along its embryogenic pathway.

Ectopic thyroid (ET) gland is a rare occurrence. It occurs in 1 per 100,000 to 300,000 individuals.1 Lingual ectopic thyroid gland accounts for 90% of the reported cases2 while extra-lingual ectopic thyroid is less frequently encountered. Cases of ectopic thyroid tissue adjacent to the esophagus, heart, aorta and pancreas have also been described. We would like to share a case of rare occurrence of ectopic thyroid gland at the right axilla presented to our center.

CASE REPORT
Madam H, a 56 years old lady presented to the Hospital Sultanah Nora Ismail (HSNI), Johor Malaysia in April 2020 for right axillary mass. She was then referred by the surgical team for optimization of her thyroid function test in September 2020.

On further history, she first noticed the right axilla swelling about 7 years earlier. The size was initially about 4cm X 5cm, non-tender, firm in consistency with no discharges. However, the axilla mass began to increase in size gradually over the previous 1 year for which she sought medical attention in April 2020. During the period of 1 year, she noticed that she had weight loss from 80kg to 60kg. She had loss of appetite as well at the same time. There were no other constitutional symptoms or B-symptoms noted.

Ultrasonography of the neck showed no significant abnormality of the orthotopic thyroid gland. A tru-cut biopsy was performed on the right axilla mass by the surgical team in April 2020 which was in consistent with benign ectopic thyroid tissue (Figure 1).

After reviewing the histopathology examination result (HPE) of the axilla mass, Madam H underwent computerized tomography scan of neck, thorax and abdomen (CECT TAP) in September 2020 for staging and for further delineation of the axillary mass. CT scan showed an enlarged, heterogeneously mass in the right axillary region with coarse calcification within measuring 7.8cm X 7.0cm X 9.3cm (Figure 2a and 2b). There was no other axillary, mediastinal or hilar lymph node enlargement. The orthotopic thyroid gland was seen at its normal location (midline anterior neck region) and demonstrating homogenous enhancement and normal in size (Figure 2c). Both lung parenchyma were normal.

As Madam H remained clinically euthyroid during the entire period of consultations in HSNI, urgent surgical resection was not performed on her. Her condition was stable on regular T.
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Table I: Results of blood investigations taken during visits to the clinic

<table>
<thead>
<tr>
<th>Date</th>
<th>Investigation</th>
<th>Result</th>
<th>Normal Range</th>
</tr>
</thead>
<tbody>
<tr>
<td>1/10/2020</td>
<td>Free T3</td>
<td>Low</td>
<td>1.88-3.18 pg/mL</td>
</tr>
<tr>
<td>12/10/2020</td>
<td>Anti-Thyroglobulin</td>
<td>473.0 (High)</td>
<td>&lt; 115 IU/ml</td>
</tr>
<tr>
<td>12/10/2020</td>
<td>Anti-Thyroid Specific Peroxidase</td>
<td>19.10</td>
<td>&lt; 34 IU/ml</td>
</tr>
<tr>
<td>12/10/2020</td>
<td>Serum Chromogranin A</td>
<td>69</td>
<td>27-94 ng/ml</td>
</tr>
</tbody>
</table>

Fig. 1: Histopathological examination shows thyroid follicles of varying sizes (a) with lumen containing colloid. The thyroid follicles are lined by simple cuboidal epithelium having uniform, rounded and basally located nuclei. There are no nuclear features of papillary thyroid carcinoma TTF-1 and thyroglobulin immunoreactivity (b-c) to support thyroidal origin.

Fig. 2: Axial (a) and coronal (b) contrast-enhanced CT images demonstrating enlarged heterogenous ectopic mass in the right axillary region (asterisk) with coarse calcification within (arrow head). Contrast-enhanced axial CT (c) demonstrating orthotopic thyroid gland (arrows) with homogenous enhancement, normal in size and at its normal location (midline anterior neck region).
L-Thyroxine 100mcg OD. However, she was later scheduled to undergo wide local resection of the ectopic thyroid gland on her right axilla on an elective operating date.

DISCUSSION
ET is an unusual presentation. The most common sites for an ectopic ET are lingual, thyroglossal and laryngotraheal which correlate with the anatomical descent of thyroid gland during embryogenesis. Only rare case reports exist of ET in other locations, including the chest (heart, trachea), abdomen (liver, gallbladder, pancreas) and pelvis (vagina). The aetiology of this abnormality is not fully understood. However, genetic factors and mutations in the regulatory genes expressed in the developing thyroid gland have been implicated in human thyroid ectopy. ET gland has risk of malignant transformation. The risk involved is less than 1% and almost all are papillary thyroid carcinomas. At the same time, radiation and obesity can increase risk of malignant transformation in ectopic organs including thyroid glands. Radiological imaging studies such as ultrasound, CT scan and Magnetic Resonance Imaging (MRI) may be helpful in knowing the extension of ET gland, but the best diagnostic test for the gland is thyroid scanning with technetium-99m.

A thyroid mass usually demonstrates high CT density on plain scan due to the iodine content and homogeneous enhancement after intravenous contrast injection. An ET tissue is typically identical in appearance to orthotopic thyroid tissue, a well-circumscribed homogeneous mass with increased attenuation. However, our patient demonstrated heterogeneous appearance of the right axillary mass, thus malignant transformation or axillary nodal metastasis were suspected initially. This dilemma was solved by tru-cut biopsy HPE reporting as benign thyroid gland. At the same time, serum chromogranin A was in the normal range and this has ruled out possible neuroendocrine tumour involvement.

A case report of benign ET tissue in the left supraclavicular has been reported, having similar CT findings of axillary mass in our patient. CT scans in our case was also vital to provide a detailed picture of masses relation to the other structures of this region to help in pre-operative plan in the future. Scintigraphy offers a useful and effective role to differentiate ET gland from other tumours especially deep-seated tumour such as in the mediastinum where procurement of tissue samples requires deep invasive surgical method. However, Madam H’s axilla mass was superficially located and thus superficial percutaneous approach was sufficient to obtain the required biopsy sample.

The treatment of ET tissue depends on factors such as mass size, symptoms, age of the patient, thyroid functional status and histological findings. In symptomatic cases, surgical resection remains the mainstay of treatment. Treatment can also be aided by hormone suppression and radioactive 131- abduction as well. Madam H received hormone replacement therapy in view of her deranged thyroid function test. As she remains euthyroid, wide local resection is being planned later in order to remove the axillary mass mainly because of the compressive symptoms.

CONCLUSION
Benign ET gland is a rare occurrence, more so in the case of lateral aberrant thyroid. Further follow up is needed as possibility of carcinomatous transformation such as papillary carcinoma arising in struma ovarii should be considered.

REFERENCES