



Benign lateral aberrant thyroid—first case report in Hong Kong

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Background: Ectopic thyroid is a rare anomaly, with a prevalence of 1 in 200,000 people. Lateral aberrant thyroid is an archaic term with the implication of malignancy. Contrary to prior understanding, this article presents a completely benign lateral aberrant thyroid, which is also the first reported case in Hong Kong.

Case Description: A 53-year-old gentleman presented with a 3 cm rapidly enlarging neck mass. Initial cytological workup with fine needle aspiration (FNA) demonstrated evidence of benign thyroid tissue, with no evidence of malignancy. However, with a presumed diagnosis of a thyroid cancer with lymph node metastasis, repeated FNA and further imaging had been arranged. Computerised tomography scan of the neck depicted a well circumscribed neck mass with areas of hypodensity and presence of orthotopic thyroid gland. Final pathology obtained from neck mass excision and hemi-thyroidectomy revealed the diagnosis of a benign ectopic thyroid and an incidental finding of a T1b 10.1 mm papillary thyroid carcinoma with clear margin.

Conclusions: To our understanding, this is the first reported case of a completely benign lateral aberrant thyroid in Hong Kong. After reviewing and comparing with other published cases in the literature, recommended management for such patients would be an ultrasound, FNA of the neck mass as first line evaluation, followed by a staged surgical approach for definitive pathological study of the lesion.

Keywords: Thyroid; ectopic thyroid; lateral aberrant thyroid; head and neck surgery; case report

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Introduction

Lateral aberrant thyroid is an archaic terminology that refers to thyroid tissue found in the lateral neck, which is anatomically separated from the orthotopic gland. It carries the connotation of malignancy since it was thought to be metastasis from thyroid carcinoma. This has been falsified by a few scattered published cases of benign lateral aberrant thyroid, including the following, the first reported case in Hong Kong. We present this case in accordance with the CARE reporting checklist (available at <https://aot.amegroups.org/article/view/10.21037/aot-23-2/rc>).

Case presentation

A 53-year-old gentleman with hypertension and obstructive sleep apnea complained of a painless progressively enlarging neck mass for months. Latest blood test showed a normal serum level of thyroid stimulating hormone at 0.96 μ IU/mL.

There was a 3 cm firm neck mass over the right level III region, otherwise, clinical examination of the head and neck region was unremarkable. Ultrasonography showed a hypoechoic mass with smooth border, deep to the sternocleidomastoid muscle. Fine needle aspiration cytology (FNAC) showed piece of thyroid tissue with

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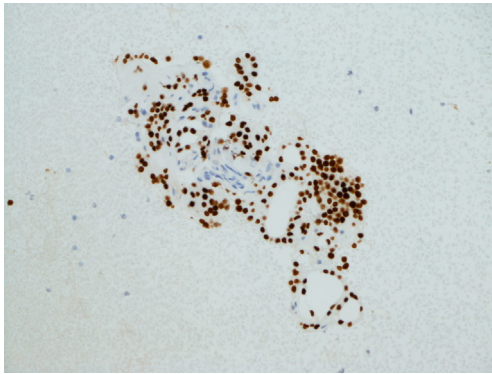


Figure 1 FNAC of the neck mass demonstrated presence of follicular cells, which are positive for TTF-1, immunohistochemically (TTF-1, $\times 200$). FNAC, fine needle aspiration cytology; TTF-1, thyroid transcription factor-1.

colloid-containing thyroid follicles. These follicular cells are positive for thyroid transcription factor-1 (TTF-1) immunohistochemically (*Figure 1*).

Computerised tomography (CT) scan of the neck depicted a well circumscribed neck mass with areas of hypodensity, which could be the result of necrosis or cystic change (*Figures 2,3*). There were several hypodense nodules, less than 1 cm in diameter over both lobes as shown by arrows. Orthotopic gland was otherwise normal in size.

In view of the progressively increasing neck mass, the patient was counselled for further imaging with positron emission tomography (PET)-CT in search of primary or definitive surgical treatment for pathological diagnosis. The patient opted for surgery. Meanwhile, further FNAC attempts were performed to the neck and to the ipsilateral thyroid respectively. The former one showed normal thyroid tissue whereas the latter was non-diagnostic.

Right neck mass excision and right hemi-thyroidectomy

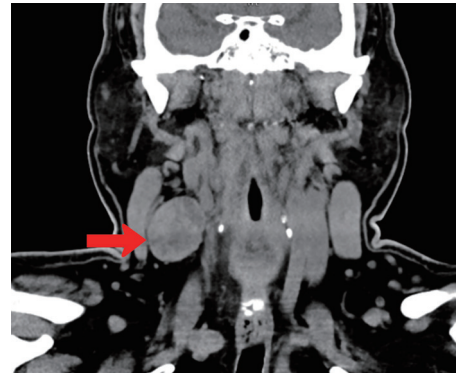


Figure 2 Coronal view: $3.0 \times 3.5 \times 4.1 \text{ cm}^3$ soft tissue mass over right level III with areas of hypodensity within, could be area of cystic change or necrosis (red arrow).

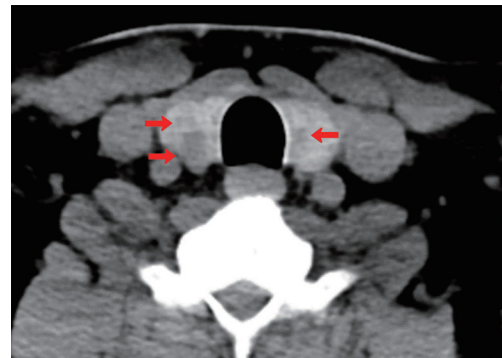


Figure 3 Sagittal view: several hypodense nodules (up to 0.7 cm) seen in both lobes of thyroid (red arrows). Thyroid gland is otherwise normal in size.

was scheduled. Intraoperatively, the right neck mass (*Figures 4,5*) can be entirely separated from the thyroid gland. Frozen section of the neck mass came back with evidence of normal thyroid tissue and a benign lymph node. Additionally, there was a firm nodule over the right thyroid lobe.

Normal thyroid tissue is demonstrated in the excised neck mass specimen (*Figure 6*). Pathology thus, confirmed the diagnosis of benign ectopic thyroid in the lateral neck region. There was an incidental T1b 10.1 mm papillary thyroid carcinoma with no lympho-vascular permeation or extra-thyroidal extension. Clear margin was obtained. Histologically, nuclear features of papillary thyroid carcinoma, with nuclear grooves and nuclear pseudo-inclusions are readily seen (*Figure 7*).

All procedures performed in this study were in accordance with the ethical standards of the institutional

Highlight box

Key findings

- Benign lateral aberrant thyroid does exist.

What is known and what is new?

- Diagnosis of a benign lateral aberrant thyroid, albeit rare, should be kept in mind when approaching and evaluating a neck mass.

What is the implication, and what should change now?

- USG + FNAC are recommended to be done for evaluation.
- Staged surgery is the preferred treatment option.

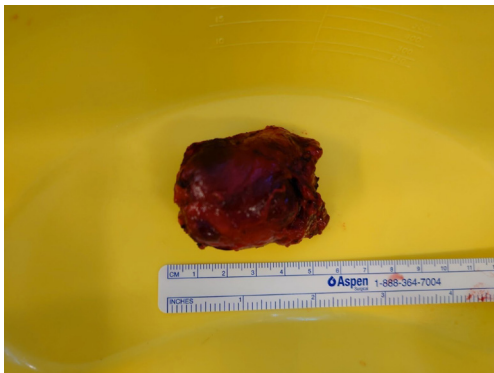


Figure 4 Intraoperative specimen photo of the neck mass (1st view).



Figure 5 Intraoperative specimen photo of the neck mass (2nd view).

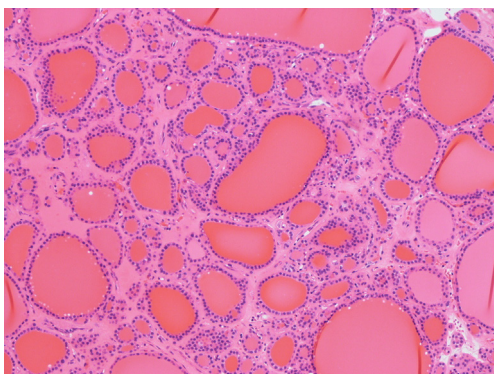


Figure 6 Presence of normal thyroid tissue is demonstrated in the excised neck mass specimen (H&E, ×100). H&E, hematoxylin and eosin.

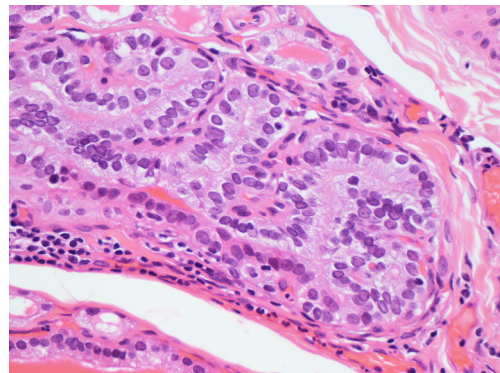


Figure 7 Incidental finding of a microscopic papillary thyroid carcinoma is present in the native thyroid. Nuclear features of papillary thyroid carcinoma, with nuclear grooves and nuclear pseudoinclusions are readily seen (H&E, ×400). H&E, hematoxylin and eosin.

and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal. In addition, the research has obtained approval from the cluster research ethics committee (REC Ref No. NTWC/REC/22084). Notice is also ready for review.

Discussion

Ectopic thyroid is a developmental abnormality, with a prevalence of 1 in 200,000 people. Most often, they are asymptomatic. Occasionally, patients may present with hypothyroidism and neck mass. Embryologically, thyroid gland is derived from a large median anlage and 2 lateral anlages. Lateral aberrant thyroid occurs when the lateral anlage fails to fuse with the median anlage at the 7th week of gestation. Lateral aberrant thyroid accounts for only 2% of all ectopic thyroid cases, yet they are generally perceived and treated as malignancy. There is one paper without cross referencing (Prado *et al.* 2012) (1), quoting a 12% rate of malignancy in lateral aberrant thyroid, which may underestimate the exact incidence because usually only rare benign cases are published.

A thorough review on the topics of benign lateral aberrant thyroid is summarised (*Table 1*). Different treatment plans were offered by respective authors, from medical treatment using levothyroxine supplementation to extensive total thyroidectomy with central and lateral neck

Table 1 Literature review on benign lateral aberrant thyroid

References	Clinical exam	Management	Pathology of the thyroid (+/- LN)		Remarks
			Ectopic	Orthotopic	
Prado <i>et al.</i> 2012 (1)	2 cm solid	Medical treatment with levothyroxine supplementation	N/A	N/A	Confirmed by 99mTc scintigraphy, followed up with FNAC
Otondi <i>et al.</i> 2020 (2)	4.3 cm rubbery	Total thyroidectomy and neck mass excision	MNG	MNG	FS sent: benign thyroid tissue
Liu <i>et al.</i> 2022 (3)	2 cm cystic	Total thyroidectomy, right lateral & central neck dissection	Benign (cysts)	MNG, 45 LNs -ve	FS of neck mass: no malignancy
Huang <i>et al.</i> 2021 (4)	N/A	Total thyroidectomy & bilateral central neck dissection	Benign	PTC, 3 LNs +ve	Bilateral PTC noted in USG and FNAC, postop RAI given

LN, lymph node; N/A, not applicable; FNAC, fine needle aspiration cytology; MNG, multinodular goitre; FS, frozen section; PTC, papillary thyroid carcinoma; USG, ultrasound-guided; RAI, radioactive iodine therapy.

dissection. Noteworthy is that a benign lateral aberrant thyroid in itself does not exclude presence of malignancy in the orthotopic thyroid gland. In fact, orthotopic thyroid gland is absent in 70% cases (5). As such, the role of preoperative ultrasound-guided (USG) would be of paramount importance, to search for primary malignancy, identify co-existing thyroid disorders, and to review the indication of thyroxine post-operatively.

To avoid a diagnostic dilemma, a staged surgery would be favoured. Pathology can be reviewed following an *en-bloc* excision of the neck mass. If malignancy is confirmed, then a decision to proceed with an ipsilateral or total thyroidectomy with neck dissection can be discussed with patients. In addition to formulating diagnosis without repeated FNAC sessions as workup or during follow up, it also helps to avoid the operative risk of extensive surgical resection including neck dissection and in the same time, negate the need for lifelong thyroxine and/or calcium supplements.

Apart from the treatment of lateral aberrant thyroid, the proposed staged surgical approach can also be applied to management of thyroid ectopia in any part of the body, in gallbladder (6), anterior mediastinum (7), and even in the heart, in the right ventricle (8). Usually asymptomatic and noted as an incidental finding, thyroid ectopia may present as a growing lump with pressure symptoms. Imaging alone therefore has limited use. Regal *et al.* performed a midline partial sternotomy with *en-bloc* excision of a 5 cm mediastinal mass for diagnostic confirmation (7). On the other hand, Comajuan *et al.* also carried out an exploratory cardiac surgery with a tricuspid valve replacement after excision of an intracardiac mass (8). Histopathology eventually

confirmed the diagnosis of ectopic thyroid tissue without evidence of malignancy in both cases. Subsequent workups in these cases included a targeted ultrasound of thyroid and thyroid function test, which was normal. Not surprisingly, the differential of thyroid ectopia was not included in both cases. Overall, thyroid ectopia indeed is a rare disease but should well be considered in the list of differential diagnosis. Preoperative ultrasound of the thyroid gland would facilitate the postoperative care of a patient when an ipsilateral or total thyroidectomy has to be discussed if malignancy is eventually confirmed. Overall, the proposed staged surgical approach could be applied in any cases of thyroid ectopia.

Conclusions

This case highlights the presence of a completely benign lateral aberrant thyroid. USG and FNAC are undeniably two important investigations that should be utilised preoperatively. A staged treatment approach is preferred to avoid potential surgical complications and the need for lifelong medication.

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Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at <https://aot.amegroups.org/article/view/10.21037/aot-23-2/rc>

Peer Review File: Available at <https://aot.amegroups.org/article/view/10.21037/aot-23-2/prf>

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at <https://aot.amegroups.org/article/view/10.21037/aot-23-2/coif>). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal. In addition, the research has obtained approval from the cluster research ethics committee (REC Ref No. NTRWC/REC/22084). Notice is also ready for review.

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