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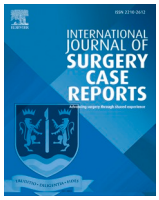

Case report

Lateral neck ectopic papillary thyroid carcinoma: A rare case report

Miray Ibrahim *, Ram Attaf, Mohammed Alhaj Saleh, Hazar Najjoum

Faculty of Medicine, Al Andalus University for Medical Sciences, Tartous, Syria

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ABSTRACT

Introduction: Ectopic thyroid tissue (ETT) occurs due to abnormal embryogenesis of the thyroid gland. Ectopic thyroid tissue is susceptible to all the processes that affect normal thyroid tissue, including malignant transformation. Malignancy of ectopic thyroid tissue is known as the exceedingly rare phenomenon of ectopic thyroid carcinoma (ETC).

Presentation of case: A 97-year old female presented with an ulcerated, hemorrhagic lateral neck mass that had been present for years prior to her visit. The mass was initially misdiagnosed as a haemangioma. During the surgical procedure, the operating surgeons discovered hallmark signs pointing towards malignancy. Histopathology of a biopsy taken from the mass confirmed the diagnosis of ectopic thyroid carcinoma (ETC).
The patient was discharged in good condition and did not require further intervention.

Discussion: The presence of ectopic thyroid carcinoma in the lateral neck is an extremely scarce finding. Medical literature details multiple modalities of diagnosis. We discuss similar cases and provide a comparison of currently applied diagnostic and therapeutic measures.

Conclusion: Thus far, no consensus has been reached regarding the optimal treatment or diagnosis of ectopic thyroid cancer, mainly due to their scarcity within the literature. As with most malignancies, early diagnosis is of great importance in order to ensure better outcomes. Individualised treatment options remain the cornerstone of the treatment of ETCs. More research is needed to outline practical tests to calibrate treatment in future cases.

1. Introduction

Ectopic thyroid tissue (ETT) is a rare condition in which thyroid tissue appears in locations other than the normal loculation in the anterior neck, between the C5 and T1 vertebrae. It occurs due to the abnormal embryogenesis of the gland, most commonly pertaining to the incomplete migration of cells from the endoderm [1].

ETT is a rare condition in itself, considering that it occurs in 1 in every 100,000 to 300,000 people [2] and mostly in young females [3]. However, developing ectopic thyroid carcinoma, defined as malignancy of the ectopic thyroid tissue, is even more rare, accounting for ~1 % of all ETT cases [1].

We report a case of a female with a lateral neck mass, later diagnosed as ectopic thyroid carcinoma. This work is compliant with the SCARE Criteria [4].

2. Presentation of case

A 97-year-old female consulted for a uniformly growing mass in the right lateral region of her neck for 25 years. The only significant symptom was neck movement restriction. The patient had no chronic diseases, nor did she have a habit of smoking or alcohol consumption. She did not have prior irradiation to the neck, and her family history is negative for thyroid cancer.

Upon physical examination, a firm, nontender, immobile, hemorrhagic, ulcerative, smooth mass was shown (Fig. 1A(Additionalfile1.pdf)). It was located at level V of the neck, posterior to the sternocleidomastoid muscle, and superior to the clavicle (Fig. 1B(Additionalfile1.pdf)). Otherwise, neck examination was nonsignificant, revealing no cervical lymph node adenopathy or any notable findings. Physical examination of other systems was normal. Thyroid functional tests TSH, T3, and T4 were normal. TSH by only 2.1 mIU/L (normal range of TSH levels in adults is between 0.4 and 4.0 mIU/L), T3 was 110 ng/dL and T4 was 5.5 µg/dL (normal range of T3: 100 to 200 ng/dL, normal range of

* Corresponding author.
E-mail addresses: mirayibr06@gmail.com (M. Ibrahim), ram.f.attaf@gmail.com (R. Attaf), hajsaleh.mohammed@gmail.com (M. Alhaj Saleh), hazarnajjoum@gmail.com (H. Najjoum).

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T4: 5.0 to 12.0 μg/dL).

Neck ultrasonography revealed a significant mass with microcalcifications and increased vascularity which, in turn, required additional investigation with CECT to accurately characterise the anatomical relationship of the mass with the surrounding tissue. CECT demonstrated a 15x12cm mass with high angiogenesis within the lateral neck and negative metastasis to local nodes. Moreover, the mass was primarily supplied by branches of the superior thyroid artery.

For further diagnosis, a fine needle aspiration biopsy (FNAB) was performed. The aspirate predominantly contained extravasated blood with scarce characteristic features on microscopy; an abundance of vessel proliferation, unclear atypia, fibrin, and granulation tissue. The atypical nonspecific findings plus the clinical features led to an initial diagnosis of haemangioma.

According to various aspects, including the patient's overall status and age, tumour site and size, and treatment goals, the chosen method of treatment was a curative *en bloc* surgery for the primary tumour. A falciform incision was deepened through the platysma muscle with a 1.5 cm margin. The subplatysmal flaps were raised, and vital structures were identified and preserved, including the eleventh cranial nerve and the internal jugular vein. Identifying and preserving the superior thyroid artery was an essential part of the procedure, since it was the main artery supplying the mass (Fig. 1C(Additionalfile1.pdf)).

A rapid intraoperative pathological examination was required; thus, a specimen was taken to perform a frozen section (FS). The outcome of the pathological analysis was papillary thyroid carcinoma (PTC) due to dysplasia in ectopic thyroid tissue. Pathological characteristics included the presence of psammoma bodies, which highly indicate PTC (Fig. 2A (Additionalfile2.pdf)). Lots of cells with Orphan Annie-eyed clear nuclei were found, which further supported the diagnosis (Fig. 2B(Additionalfile2.pdf)). Other features included tall, branching papillae lined with cells displaying all forms of atypia, such as irregular chromatin patterns and membranes, and nuclear pseudoinclusions (Fig. 2C(Additionalfile2.pdf)). In addition to that, the eutopic thyroid was pathologically studied to make sure no malignancy was present to rule out the diagnosis of metastasis.

The surgery resulted in the resection of both the tumour and the sternocleidomastoid muscle, as well as the dissection of level V lymph nodes. During surgery, there was no connection to the right lobe of the thyroid, and there were no complications. The incision was closed in layers. The patient received adequate postoperative care and did not undergo adjuvant therapy. She was later discharged in good overall health.

3. Discussion

Ectopic thyroid tissue (ETT) is the end result of the failure in migration of the thyroid diverticulum from the foramen caecum to its final position anterior to the trachea [1]. The prevalence of ETT is approximately 1 in 100,000 to 300,000; however, a prevalence of 7–10 % has been reported in autopsy findings, which suggests likely underdiagnosis [2].

ETT is most commonly found in the lingual location, but can be found anywhere along the path of the thyroid gland's descent. Rarer locations include the heart, adrenal glands, pancreas, and even skin [1]. 1–3 % of ETT cases are localised in the lateral cervical region [5]. Lateral neck thyroid ectopias can occur due to failure in the fusion of the lateral and median anlagen [7].

Malignancy of the ectopic thyroid tissue is exceedingly rare, with a reported incidence of <1% [8]. Histological subtypes of ectopic thyroid cancer include papillary cell carcinoma, the most common type, mixed papillary, and Hurthle cell tumors [6].

Several factors, such as separate blood supply to the ectopic gland, negative history of cancer and neck surgeries, and a normal or absent eutopic thyroid gland, support the diagnosis of ectopic thyroid carcinoma, in contrast to a metastasis from the eutopic gland [2]. In our case, our patient had no history of cancer, neck surgery, or any symptoms outside of neck movement restriction due to the increasing size of the mass. The lack of any fibrous bands connecting the ectopic tissue and the thyroid, as well as its location in level V, not levels II and III, further cements the independent nature of the ectopic tissue carcinoma [9]. Thyroid function tests were normal.

Because of the mostly asymptomatic nature of this pathology, a preoperative diagnosis is difficult to confirm. Imaging choices include scintigraphy, computed tomography (CT), and magnetic resonance imaging (MRI). Commonly, the first step in imaging is performing an ultrasound. It is noteworthy that the normal thyroid gland can mask the presence of ectopy by uptaking most of the radionuclide. CT and MRI aid in accurately determining the extension of the mass and its relation to surrounding structures, which is valuable for pre-surgical planning [9]. Accordingly, our patient underwent a CECT prior to surgical intervention. An FNAB provides critical information on the nature of the mass. The main immunomarker for thyroid tissue is thyroglobulin (Tg), as well as mRNA in needle washout. It is worth mentioning that FNAB struggles as a diagnostic method for masses of a cystic nature [10]. Furthermore, a high false negative rate was reported for FNAB in cancer of the thyroglossal duct cyst [2]. In the present case, the patient presented with a highly vascularised, ulcerated lateral neck mass. This could have played a role in the misdiagnosis of a haemangioma, as FNAB cytology showed vascular endothelium and extravasated blood. A study by Cabibi et al.

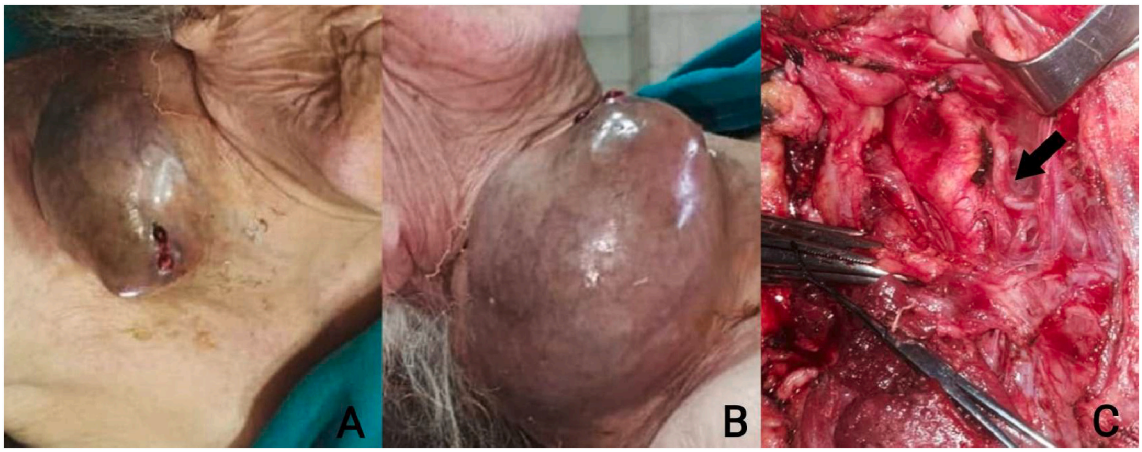

Fig. 1. A. An ulcerative and smooth-surfaced mass located on the right side of the patient's neck. B. Lateral view of the mass that demonstrates its location above the clavicle and posterior to the sternocleidomastoid muscle. C. The superior thyroid artery (black arrow) that was mainly supplying the mass.

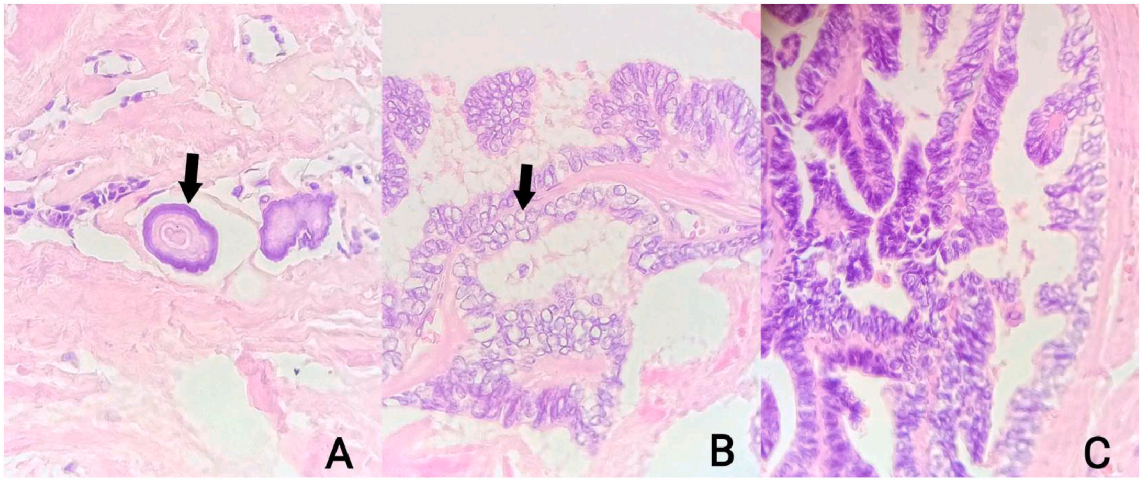

Fig. 2. A. A psammoma body shown on histopathological examinations (black arrow). B. Orphan Annie-eyed clear nuclei (black arrow). C. Branching papillae lined with cells that show all forms of atypia, like irregular chromatin patterns and membranes, and nuclear pseudoinclusions.

explored the role of the immunohistochemistry markers cytokeratin-19, HBME-1, and galectin-3 in differentiating between lateral neck metastases of papillary thyroid carcinoma and primary carcinogenesis within ectopic thyroid tissue. The study compared morphologically normal-appearing lateral neck thyroid tissue between two groups of patients. All three biomarkers were positive in patients with eutopic thyroid tumors, confirming the metastatic nature of these lateral neck masses. On the other hand, cancer-free eutopic thyroid glands tested negative for all three biomarkers, which establishes the lateral neck masses as growths independent of the eutopic thyroid. In our case, due to limited resources, such tests were simply not at our disposal. The aforementioned study highlights the necessity of future, larger-scale studies that further explore the role of these biomarkers in differentiating metastases from primitive cancers while avoiding further surgical interventions, namely total thyroidectomy [11].

To this date, there has been no definitive optimal surgical mode of treatment of lateral neck ETC. It can be difficult to assess whether the extrathyroid mass is a metastasis of an underlying cancer of the orthotopic thyroid gland or a separate, independent mass. The diagnosis of an ETC warrants a thorough evaluation of the orthotopic thyroid gland for any possible malignancy. The most important factor in determining the need for total thyroidectomy is the presence or absence of concomitant carcinogenesis in the orthotopic thyroid gland, which can be visualised through imaging modalities such as ultrasonography and CT scans [1]. However, the final diagnostic modality remains histological examination of the orthotopic thyroid tissue. In a study by Lee et al., a 24-year-old euthyroid female with a right lateral neck mass with lymphadenopathy underwent total thyroidectomy with selective lymph node dissection due to suspicion of metastatic papillary thyroid cancer. However, histopathology of the thyroid gland only revealed chronic lymphocytic thyroiditis without any signs of malignancy [12]. In another study by Singh et al., a 53-year-old male was diagnosed with metastatic papillary thyroid carcinoma based on FNAB findings from a lateral neck mass. The patient underwent a subtotal thyroidectomy with radical neck dissection. Nonetheless, histological analysis of the eutopic thyroid showed no signs of malignancy [13]. This sheds light on the need for developing a highly individualised treatment strategy and, perhaps, calls for less invasive methods for determining the presence or absence of cancer in the eutopic thyroid gland. In our case, the patient's orthotopic gland showed no features suggestive of malignancy on both ultrasonography and CT scanning, the latter showed no regional lymphadenopathy, which favoured a less invasive surgical approach. Postoperatively, FNABs from multiple regions of the eutopic gland with thorough histological examination revealed no foci of malignancy.

4. Conclusion

The rarity of ETC accounts for the difficulty of diagnosis and potential misdiagnosis and underdiagnosis, especially with the risk of false negatives in diagnostic procedures. Even with the low prevalence, further tests like immunomarkers and scintigraphy should be conducted. Moreover, more research is needed to establish concrete testing methodologies to aid physicians in making suitable treatment decisions.

List of abbreviations

CECT	Contrast-Enhanced Computed Tomography
CT	Computed Tomography
ETC	Ectopic Thyroid Carcinoma
ETT	Ectopic Thyroid Tissue
FNAB	Fine Needle Aspiration Biopsy
FS	Frozen Section
HBME-1	Human Bone Marrow Endothelial Cell Marker-1
MRI	Magnetic Resonance Imaging
PTC	Papillary Thyroid Carcinoma
TG	Thyroglobulin

Author contribution

Miray Ibrahim contributed to proof-reading, final drafting, and figures interpretations.

Mohammed Alhaj Saleh reported the pathologic findings, operation notes, and the clinical findings.

Hazar Najjoum contributed to literature search and reviews.

Ram Attaf contributed to the discussion of the case.

All authors contributed to writing equally and read and approved the final manuscript.

Consent

Written informed consent was obtained from the patient's next of kin for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

We point out that it is not necessary in our institution to submit or get an approval by an ethics committee. The institution to which we belong

is Al Andalus University for Medical Sciences, Tartous, Syria.

Guarantor

Miray Ibrahim.

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Conflict of interest statement

The authors declare that they have no conflicts of interest.

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References

[1] J. Klubo-Gwiezdzinska, R.P. Manes, S.H. Chia, et al., Ectopic cervical thyroid carcinoma—review of the literature with illustrative case series, J. Clin. Endocrinol. Metab. 96 (9) (2011) 2684–2691, https://doi.org/10.1210/jc.2011-0611.

[2] G. Noussios, P. Anagnostis, D.G. Goulis, D. Lappas, K. Natsis, Ectopic thyroid tissue: anatomical, clinical, and surgical implications of a rare entity, Eur. J. Endocrinol. 165 (3) (2011) 375–382, https://doi.org/10.1530/EJE-11-0461.

[3] C. Sohrabi, G. Mathew, N. Maria, et al., The SCARE 2023 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, Int. J. Surg. 109 (5) (2023) 1136–1140, https://doi.org/10.1097/JS9.0000000000000373.

[4] A. Toso, F. Colombani, G. Averono, P. Aluffi, F. Pia, Lingual thyroid causing dysphagia and dyspnoea. Case reports and review of the literature. Acta otorhinolaryngologica Italica : organo ufficiale della Societa italiana di otorinolaringologia e chirurgia cervico-facciale. 29 (4) (2009) 213–217. https://www.ncbi.nlm.nih.gov/pmc/articles/PMC2816370/.

[5] H. Prado, A. Prado, B. Castillo, Lateral ectopic thyroid: a case diagnosed preoperatively, Ear Nose Throat J. 91 (4) (2012) E14–E18, https://doi.org/10.1177/014556131209100417.

[6] B.C. Shah, C.S. Ravichand, S. Juluri, A. Agarwal, C.S. Pramesh, R.C. Mistry, Ectopic thyroid cancer, Ann. Thorac. Cardiovasc. Surg. 13 (2) (2007) 122–124.

[7] G. Guerra, M. Cinelli, Massimo Mesolella, et al., Morphological, diagnostic and surgical features of ectopic thyroid gland: a review of literature, Int. J. Surg. 12 (12) (2014) S3–S11, https://doi.org/10.1016/j.ijsu.2014.05.076.

[8] Y. Agosto-Vargas, M. Gutiérrez, J.H. Martínez, et al., Papillary thyroid carcinoma: ectopic malignancy versus metastatic disease, Case Rep Endocrinol. 2017 (2017) 9707031, https://doi.org/10.1155/2017/9707031.

[9] D.A. Zander, W.R. Smoker, Imaging of ectopic thyroid tissue and thyroglossal duct cysts, Radiographics 34 (1) (2014) 37–50, https://doi.org/10.1148/rg.341135055.

[10] R. Bellantone, C.P. Lombardi, M. Raffaelli, et al., Management of cystic or predominantly cystic thyroid nodules: the role of ultrasound-guided fine-needle aspiration biopsy, Thyroid 14 (1) (2004) 43–47, https://doi.org/10.1089/105072504322783830.

[11] D. Cabibi, M. Cacciatore, C. Guarnotta, F. Aragona, Immunohistochemistry differentiates papillary thyroid carcinoma arising in ectopic thyroid tissue from secondary lymph node metastases, Thyroid 17 (7) (2007) 603–607, https://doi.org/10.1089/thy.2007.0063.

[12] Ainee Kyrstelle Lee, P. Marie, P. Siy, Dahlia Teresa Argamosa, Ectopic papillary thyroid carcinoma presenting as right lateral neck mass: a case report, Journal of the ASEAN Federation of Endocrine Societies. 37 (1) (2022) 103–106, https://doi.org/10.15605/jafes.037.01.18.

[13] V. Singh, T. Srinivas, S. Bhat, S. Goel, Massive lateral neck mass: aberrant ectopic thyroid malignancy, BMJ Case Rep. 14 (5) (2021) e241451, https://doi.org/10.1136/bcr-2020-241451.