

Contents lists available at [ScienceDirect](https://www.sciencedirect.com)

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

AGGRESSIVE PAPILLARY CARCINOMA OF THE LATERAL ABERRANT THYROÏDE: A CASE REPORT AND REVIEW OF THE LITERATURE

Aggressive papillary carcinoma of the lateral aberrant thyroïde: A case report and review of the literature

Ahmed Brahim Ahmedou^{a,*}, Chaker Kaoutar^a, Youssef Oukessou^a, Redallah Abada^a, Rouadi Sami^a, Roubal Mohamed^a, Mahtar Mohamed^a, Tolba CSA Karkouri Mehdi^b

^a ENT Department, Face and Neck Surgery, Hospital August, 20'1953, University Hospital Centre IBN ROCHD, Casablanca, Morocco

^b Pathology Department, Centre IBN ROCHD, Casablanca, Morocco

ARTICLE INFO

Article history:

Received 14 June 2020

Received in revised form

11 September 2020

Accepted 11 September 2020

Available online 22 September 2020

Keywords:

Lateral aberrant thyroïde

Papillary carcinoma

Pseudoaneurysm

ABSTRACT

INTRODUCTION: Ectopic thyroid tissue can be found in many sites. Ectopic thyroid tissue may also be involved in the same processes as normal thyroid gland. These processes include tumors, inflammation and hyperplasia. The appearance of such tissue in rare locations may lead to diagnostic and therapeutic dilemmas.

PRESENTATION OF CCASE: We report a rare case of primary papillary carcinoma in an ectopic thyroid in the jugulocarotid region based on the bifurcation of the carotid artery, in a 62-year-old woman with history of diabetes and high blood pressure, with a normal thyroid gland.

DISCUSSION: The origin of lateral ectopic thyroid tissue is not fully understood and controversial. The lateral localisation is a rare entity and debated extensively in the literature.

CONCLUSION: Though rare, the possibility of an ectopic thyroid carcinoma must always be considered by the surgeon in cases of a pathological mass in the neck.

© 2020 Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

1. Introduction

This work has been reported in line with the SCARE criteria and cite the following paper: Agha RA, Borrelli MR, Farwana R, Koshy K, Fowler A, Orgill DP, For the SCARE Group. The SCARE 2018 Statement: Updating Consensus Surgical Case Report (SCARE) Guidelines, International Journal of Surgery 2018;60:132–136.

Ectopic thyroid is defined as the presence of thyroid tissue in locations other than the normal anterior neck region. It is a rare developmental abnormality that involves aberrant embryogenesis of the thyroid gland when it migrates from the floor of the primitive foregut to its final region between the second and fourth tracheal cartilages [2]. The prevalence reported as 1 per 100 000–300 000 people, rising to 1 per 4000–8000 patients having thyroid disease [2]. Ectopic thyroid may become goitrous [3] or associated with thyroid dysfunction, hypofunction [4] or hyperfunction but malignancy is uncommon particularly primary thyroid carcinomas and still less than 1% [5,6]. On the other side however, making the difference between primary carcinoma and a metastatic carcinoma is a challenging situation.

The aim of this study is to present from our case the clinical, biological and radiological behavior of an unusual aggressiveness of a primary papillary carcinoma in an ectopic thyroid in the jugulocarotid region with pseudoaneurysm of the carotid artery, and to discuss from the literature review its management.

2. Presentation of the case

A 62-year-old woman with history of diabetes and high blood pressure, non-smoker and non-alcoholic with no history of radiation exposure or significant family history; admitted to the emergency room with a spontaneous bleeding from a right laterocervical mass, which appeared 30 years ago and which increases in size in the last 6 months. She did not show symptoms of hypothyroidism or hyperthyroidism or any other symptoms.

Physical examination revealed a mass measuring approximately 6 cm in its long axis, at the level of the jugulocarotid region, non-tender, swinging, fistulized to the skin with blood issue (Fig. 1). The rest of the clinical examination was unremarkable.

* Corresponding author at: ENT Department, Face and Neck Surgery, Hospital August, 20'1953, University Hospital Centre IBN ROCHD, Street mausolée imm 10, app 9, quartiers des hopitaux, Casablanca, Morocco.

E-mail address: a.ahmedbrahim@gmail.com (A.B. Ahmedou).



Fig. 1. The clinical aspect of the mass.

Cervical ultrasound (US) mounted a right laterocervical mass, roughly oval, with irregular contour and heterogeneous echotexture, strongly vascularized, and containing areas of necrosis and measuring 58 * 33 * 49 mm. the thyroid gland was of normal echotexture and size, notably the parotid, submandibular and sublingual glands were normal with no cervical lymphadenopathy. On preoperative cross-sectional imaging with CT+ MRI, showed a large heterogeneous enhancing lesion with ill-defined margins on the bifurcation of the carotid artery, while achieving an aspect of pseudoaneurysm (Fig. 2). No lymphnodes were involved. No invasion of the surrounding structures. The thyroid and salivary glands were normal. Results of thyroid function tests (free T3, free T4, and thyroid stimulating hormone (TSH)) were within normal range.

The patient underwent complete excision surgery with extemporaneous examination that showed tubulopapillary and vesicular thyroid papillary carcinoma developed on an ectopic thyroid nodule. Although there was no anatomical connection to the thyroid gland, a total thyroidectomy was performed during the same surgery (Fig. 3). She did not benefit from cervical lymph node dissection because there were no clinical or radiological lymphadenopathies. Histologically, the showed carcinomatous proliferation of a papillary nature, without associated lymph node tissue. It is made of tubulopapillary and follicular structure, within a dense fibrocollagenstroma, with a cubo-cylindrical coating, made of cells with moderately eosinophilic cytoplasm, and with nucleus



Fig. 2. MRI shows a heterogeneously enhancing large lesion with, while achieving an aspect of pseudoaneurysm.

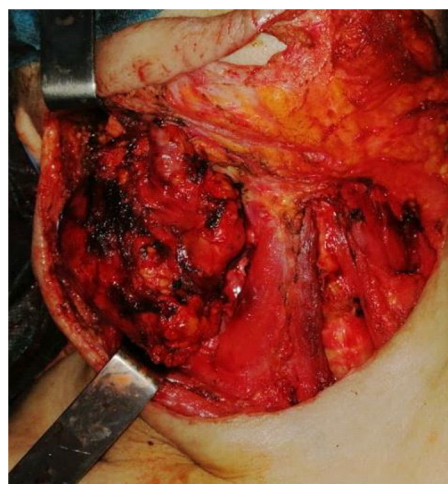


Fig. 3. Intraoperative image.

seat of cytonuclear atypia (Fig. 4). Absence of image of vascular emboli. The thyroid gland had normal tissue, notably no histological signs of thyroiditis. Instead, the patient was referred to the nuclear medicine service for evaluation with a ¹³¹Iodine whole-body scan (WBS) and possible treatment with radioiodine (RAI). The patient is on follow-up (more than 4 years) and remains well with no evidence of recurrence.

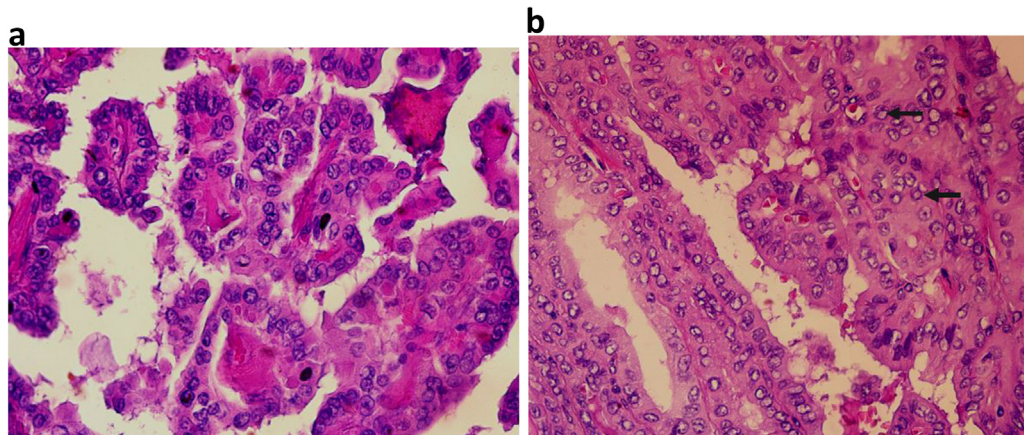


Fig. 4. (A) note here especially numerous nuclear incisions with nuclei increased in size and overlapped and containing a dusty chromatin as well as multiple small nucleoli. (B) Among the papillary nuclear criteria we note here some intranuclear cytoplasmic pseudo-inclusions (arrows), optically empty nuclei as well as an irregularity of the nuclear membrane by place.

3. Discussion

Thyroid gland embryologically is an endodermic derivative which starts to develop at the 24th day of gestation and arrives to its final location by the 7th week of gestation; originates from two different structures. During this migration, part or all of the thyroid-forming may not descend to its normal location; resulting in the appearance of ectopic tissue [7,8]. This ectopic tissue may be found anywhere from the base of the tongue to the diaphragm. Lingual, thyroglossal, laryngotracheal are the most frequent sites. Other less frequent sites are the esophagus, mediastinum, heart, adrenal glands, and pancreas [9]. The location in lateral neck to the jugular vein is controversial; this is called lateral aberrant thyroid [10] because it was thought to be metastasis from thyroid carcinoma [3,11]. The origin of lateral ectopic thyroid tissue is not fully understood and controversial. This can be explained by the fact that several disease processes can conduct detaching fragments of thyroid tissue in the neck which is not associated with lymph nodes; and it includes nodular goiter and chronic lymphocytic thyroiditis [10]. Thus some authors suppose that it originates from lateral thyroid anlagen (ultimobranchial bodies) that failed to fuse with the median anlage during caudal migration [12,13]. An example from the literature is that of Ibrahim et al., [3] where three separate ectopic thyroid masses in the lateral neck region associated to ectopic goitre.

Most of patients having ectopic thyroid do not present symptoms; they become symptomatic only if endocrine dysfunction and/or with increase size. Regarding age, there is two statistical peaks of ages which are 12.5 and 50 years [14]. The clinical examination shows characteristically a smooth margin mass soft in consistency, mobile and not-tender. It should be differentiated from thyroglossal duct cyst, epidermal cyst, lymphadenopathy, lipoma, lymphangioma, and other subcutaneous swellings and neoplasms [15].

Radionuclide thyroid imaging employing technetium-99 m pertechnetate, iodine-131 or iodine 123 is useful in the evaluation for ectopic thyroid [16] but high resolution ultrasound (US) is favoured in the initial assessment. It is non-invasive, cost-effective and does not expose patients to ionizing radiation [17]. On CT scans, ectopic thyroid tissue is seen as a homogeneous, well-circumscribed mass that, it enhances contrast after the administration of iodinated contrast [18,19]. The lateral localisation is a rare entity and debated extensively in the literature [12,13,20,21]. For this, and according to some authors lateral ectopic tissue is defined as a lateral tissue, superficial to the strap muscles without midline continuity because most of cases have been reported closely related to the strap muscles [12,13,22]. Only few cases have been in the submandibular region [12,23], jugulodigastric region [24], or within the parotid gland substance [25] but no case has been published in the division of the carotid artery, in our case as described.

The probability of malignancy in ectopic thyroid is low, less than 1%, and when it happens papillary carcinoma is the most common cancer [26,27], but most authors agree that lateral thyroid tissue is rarely benign in nature [10]. Thus aberrant thyroid tissue of the head and neck should also guide to metastatic disease from an occult primary carcinoma of the thyroid [10] specially when it is agreed that lymph node metastasis is common, distant metastases can happen in 10% of cases (28) % of cases [28]; otherwise lymph node metastases in malignant ectopic lesions are present in 30% of cases [6,28]. The differentiation between metastatic carcinoma and primary ectopic thyroid tissue is a real challenge because it is important to exclude a primary thyroid malignancy before making the diagnosis of benign aberrant thyroid tissue; this is due to the fact that well-differentiated thyroid carcinoma might metastasize even in small or occult tumors [10,28]. In our case, the histopathological study objectified the absence of lymph node tissue, and the

piece of thyroidectomy was of normal aspect without any sign of neoplasia.

Although there is no consensus regarding the optimal therapeutic strategy, due to the rarity of this entity, most authors indicate surgery depends on size and local symptoms (airway obstruction, dysphagia, and dysphonia), complications as (ulceration, bleeding, cystic degeneration, or malignancy), as well as on other parameters, such as patient's age, functional thyroid status [3,13]. In this attitude, despite the idea of a total thyroidectomy remains a "myth" as a radical intent of surgery, thyroid remnants are almost always present. Salvatori et al. reported an incidence of remnants tissue in 6.9 % of the patients having undergone total thyroidectomy for carcinoma [30,31]. Higher doses of radioiodine may be required for size reduction [32]. Ablative radioiodine should be avoided in children and young adults, due to deleterious side effects on the gonads and other organs [29,33].

4. Conclusion

Thyroid cancer arising from ectopic tissue remains a rare entity. The possibility of an ectopic thyroid cancer in the setting of a normal thyroid gland should be considered as a differential diagnosis in cases of an identified neck mass. This case report demonstrates that a normal thyroid gland does not exclude the presence of thyroid carcinoma in an ectopic tissue.

Declaration of Competing Interest

The authors report no declarations of interest.

Source of funding

None.

Ethical Approval

I declare on my honor that the ethical approval has been exempted by my establishment.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Author contribution

Ahmed Brahim Ahmedou: Corresponding author writing the paper.

Chaker Kaoutar: writing the paper.

Youssef Oukessou: study concept.

Sami Rouadi: study concept.

Reda Abada: study concept.

Mohammed Roubal: correction of the paper.

Mohamed Mahtar: correction of the paper.

Cheikh Sidahmed Tolba: data analysis.

KARKOURI Mehdi: data analysis.

Registration of Research Studies

researchregistry2464.

Guarantor

DR AHMED BRAHIM AHMEDOUr Ahmed Brahim Ahmedou.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Acknowledgements

Our sincere gratitude to our Professors and to all the staff of the ENT Department, Face and Neck Surgery, August Hospital, 20th 1953, IBN ROCHD University Hospital Center, Casablanca, Morocco.

References

- [1] Md Felice, Rd Lauro, Thyroid development and its disorders: genetic and molecular mechanisms, *Endocr. Rev.* 25 (2004) 722–746.
- [2] F. Babazade, H. Mortazavi, H. Jalalian, E. Shahvali, Thyroid tissue as a submandibular mass: a case report, *J. Oral Sci.* 51 (2009) 655–657, <http://dx.doi.org/10.2334/josnusd.51.655>.
- [3] Na Ibrahim, Ma oludara, Lateral cervical ectopic thyroid masses with eutopic multinodular goiter: an unusual presentation, *Hormones (Athens)* 8 (2009) 150–153.
- [4] Na al-Jurayyan, Mi El-desouki, Transient Iodine organification defect in infants with Ectopic thyroid glands, *Clin. Nucl. Med.* 22 (1997) 13–16.
- [5] B.C. Shah, C.S. Ravichand, S. Juluri, A. Agarwal, C.S. Pramesh, R.C. Mistry, Ectopic thyroid cancer, *Ann. Thorac. Cardiovasc. Surg.* 13 (2007) 122–124.
- [6] T. Abraham, H. Schöder, Thyroid cancer—indications and opportunities for positron emission tomography/computed tomography imaging Seminars in Nuclear Medicine, 41, 2011, pp. 121–138, no. 2, Elsevier.
- [7] 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 (2011) 375–382.
- [8] J. Klubo-Gwiedzinska, R.P. Manes, S.H. Chia, K.D. Burman, N.A. Stathatos, Z.E. Deeb, L. Wartofsky, Ectopic cervical thyroid carcinoma – review of the literature with illustrative case series, *JCEM* 96 (2011) 2684–2691.
- [9] G. Lianos, C. Bali, V. Tatsis, et al., Ectopic thyroid carcinoma. Casereport, *Giornale di Chirurgia* 34 (4) (2013) 114–116.
- [10] Haruko A. Kuffner, Barry M. McCook, Rajendiran Swaminatha, Eugene N. Myers, Jennifer L. Hunt, Controversial ectopic thyroid: a case report of thyroid tissue in the Axilla and benign total thyroidectomy, *Thyroid* 15 (9) (2005), © Mary Ann Liebert, Inc.
- [11] J.Y. Choi, J.H. Kim, Case of an ectopic thyroid gland at the lateral neck masquerading as a metastatic papillary thyroid carcinoma, *J. Korean Med. Sci.* 23 (2008) 548–550.
- [12] 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.
- [13] T.P. Caccetta, A. Kumar, H. Ishak, Lateral aberrant thyroid tissue presenting as a lateral neck mass, *ANZ J. Surg.* 75 (12) (2005) 1123–1124.
- [14] R. Rahbar, M.J. Yoon, L.P. Connolly, et al., Lingual thyroid in children: a rare clinical entity, *Laryngoscope* 118 (2008) 1174–1179.
- [15] E.N. Mussak, A. Kacker, Surgical and medical management of midline ectopic thyroid, *Otolaryngol. Head Neck Surg.* 136 (2007) 870–872.
- [16] Nasiru Akamnu Ibrahim, Idowu Olusegun Fadeyibi, Ectopic thyroid: etiology, pathology and management, *Hormones* 10 (4) (2011) 261–269.
- [17] H. ohnishi, H. Sato, H. Noda, H. inomata, N. Sasaki, Color doppler ultrasonography: diagnosis of ectopic thyroid gland in patients with congenital hypothyroidism caused by thyroid dysgenesis, *J. Clin. Endocrinol. Metab.* 88 (2003) 5145–5149.
- [18] A.L. Weber, G. Randolph, F.G. Aksoy, The thyroid and parathyroid glands. CT and MR imaging and correlation with pathology and clinical findings, *Radiol. Clin. North Am.* 38 (5) (2000) 1105–1129.
- [19] C. Altay, N. Erdogan, Karasu Setal, CT and MRI findings of developmental abnormalities and ectopia varieties of the thyroid gland, *Diagn. Interv. Radiol.* 18 (4) (2012) 335–343.
- [20] H. Prado, A. Prado, B. Castillo, Lateral ectopic thyroid: a case diagnosed preoperatively, *Ear Nose Throat J.* 91 (4) (2012) E14–E18.
- [21] J.G. Batsakis, A.K. El-Naggar, M.A. Luna, Thyroid gland ectopias, *Ann. Otol. Rhinol. Laryngol.* 105 (12) (1996) 996–1000.
- [22] M.R. Pelizzo, F. Torresan, G. Grassetto, G. Briani, M.C. Marzola, D. Rubello, Imaging identifies submandibular ectopic thyroid tissue, *Clin. Nucl. Med.* 36 (8) (2011) 728–730.
- [23] R.K.N.F. Lingam, E. Nigar, A. Kalan, Chronic lymphocytic thyroiditis presenting as a jugulodigastric neck mass, *Otorhinolaryngologist* 3 (3) (2011) 118–120.
- [24] V.V. Mysorekar, C.P. Dandekar, M.R. Sreevathsa, Ectopic thyroid tissue in the parotid salivary gland, *Singap. Med. J.* 45 (9) (2004) 437–438.
- [25] A. Sahbaz, N. Aksakal, B. Ozcinar, F. Onuray, K. Caglayan, Y. Erbil, The “forgotten” goiter after total thyroidectomy, *Int. J. Surg. Case Rep.* 4 (3) (2013) 269–271.
- [26] 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.
- [27] Oscar R. Vázquez, Frieda Silva, Eduardo Acosta-Pumarejo, Maria L. Marín, Ectopic papillary thyroid cancer with distant metastasis, *Hindawi Case Rep. Endocrinol.* 2018 (2020) 4, <http://dx.doi.org/10.1155/2018/8956712>, 8956712.
- [28] A. Toso, F. Colombani, G. Averono, P. Aluffi, F. Pia, Lingual thyroid causing dysphagia and dyspnoea. Case reports and review of the literature, *Acta Otorhinolaryngol. Ital.* 29 (2009) 213–217.
- [29] J.Y. Kwak, K.H. Han, Yoon J. Hetal, Thyroid imaging reporting and data system for US features of nodules: a step in establishing better stratification of cancer risk, *Radiology* 260 (3) (2011) 892–899.
- [30] M.C. Frates, C.B. Benson, P.M. Doubilet, et al., Prevalence and distribution of carcinoma in patients with solitary and multiple thyroid nodules on sonography, *J. Clin. Endocrinol. Metab.* 91 (9) (2006) 3411–3417.
- [31] F.W. Neinas, C.A. Gorman, K.D. Devine, L.B. Woolner, Lingual thyroid. Clinical characteristics of 15 cases, *Ann. Intern. Med.* 79 (1973) 205–210.
- [32] D.J. Alderson, F.J. Lannigan, Lingual thyroid presenting after previous thyroglossal cyst excision, *J. Laryngol. Otol.* 108 (1994) 341–343, <http://dx.doi.org/10.1017/S0022215100126714>.

Open Access

This article is published Open Access at [sciencedirect.com](https://www.sciencedirect.com). It is distributed under the [IJSCR Supplemental terms and conditions](#), which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.