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

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1. Introduction

This work has been reportor neck region [1] ed 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.

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Cervical ultrasound (US) mounted a right laterocervical mass, roughly oval, with irregular contour and heterogeneous echostructure, strongly vascularized, and containing areas of necrosis and measuring 58 * 33 * 49 mm. the thyroid gland was of normal-chostructure and size, notably the parotid, submandibular and sublingual glands were normal with no cervical lymphadenopathy. On preoperative cross-sectional imaging with CT + MRL, 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 involed. 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 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.

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**Fig. 1.** The clinical aspect of the mass.

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

**Fig. 3.** Intraoperative image.

**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 detached 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 50years[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; 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 therapeutich 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 radiiodine may be required for size reduction [32]. Ablative radiiodine should be avoided in children and young adults, due to deleterious sides 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.

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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.

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Guarantor

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