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## Anaplastic carcinoma in submandibular region: A diagnostic dilemma



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## ABSTRACT

**INTRODUCTION:** Thyroid carcinoma arising in an extrathyroid area is a rare entity.

**PRESENTATION OF CASE:** We report a case of anaplastic carcinoma in the submandibular region occurring in a 70-year-old woman.

**DISCUSSION:** The location of the mass along with no evidence of primary tumor at the orthotopic thyroid gland posed a diagnostic dilemma: was this an ectopic thyroid carcinoma or rather a case of occult differentiated thyroid carcinoma metastasis that transformed to anaplastic carcinoma? Based on the histopathological findings we concluded that the tumor arised in ectopic thyroid tissue. Although the mass was completely resected, the patient died 10 months after diagnosis due to pulmonary metastases.

**CONCLUSION:** Conclusively, the possibility of ectopic thyroid tissue, with or without disease, should be considered in cases of a mass in the submandibular region.

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## 1. Introduction

Thyroid gland derives mainly from an endodermal diverticulum, which develops in the midline at the base of the tongue. This diverticulum migrates caudally, along the thyroglossal duct, which normally obliterates by the end of the eighth embryonic week. Any defect in this embryogenetic process may lead to ectopic thyroid development. This congenital disorder is usually encountered in the midline cervical region along the course of thyroglossal duct arising from the base of tongue to the lower neck. Laterally located ectopic thyroid tissue, with or without a normally located thyroid gland, is a very rare entity [1,2]. In such cases, the clinical presentation varies depending on the location of ectopic thyroid tissue [3].

Ectopic thyroid may develop the same diseases that affect the normal orthotopic thyroid gland [4]. However, primary carcinomas arising from ectopic thyroid tissue are very rare. Namely, the probability of ectopic thyroid carcinoma is less than 1% [5]. The majority of carcinomas observed in extrathyroid locations are reported to be papillary. To the best of our knowledge, very few reports of anaplastic carcinoma arising in ectopic thyroid gland have been published

to date. Here, we report on a rare case of anaplastic carcinoma located in submandibular region in a 70-year-old woman.

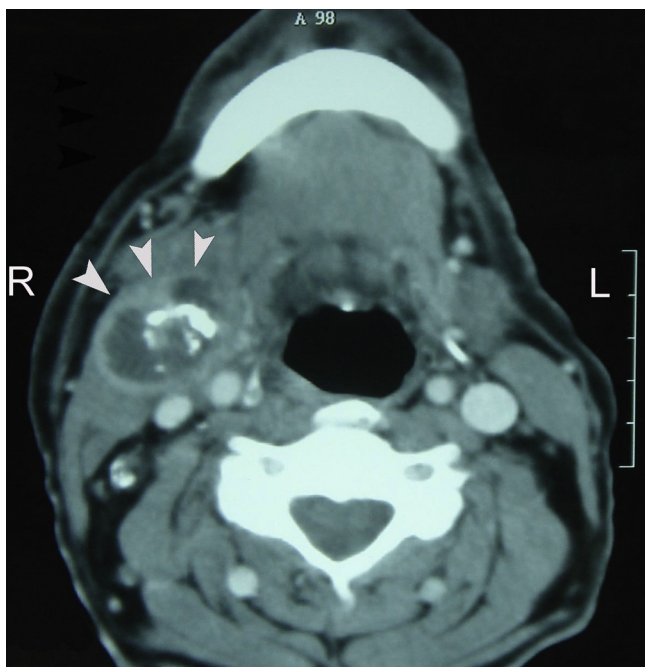
## 2. Presentation of case

A 70-year-old woman presented at the ENT outpatient department of University Hospital of Larissa complaining of a painless mass in the right submandibular region. The mass was dormant for twenty years and grew up progressively rapidly since two months ago. The patient had no prior history of head and neck radiation exposure or family history of thyroid disease or malignancy. There were no symptoms suggestive of disturbed thyroid status. Clinical examination revealed a firm level 2 mass in the right submandibular region. Flexible nasopharyngoscopy showed no mucosal lesions and normal vocal cord appearance and function. On physical examination no further palpable masses were found.

Initially an ultrasound was performed. A 54 mm solitary cystic lesion was shown with irregular margins in the right submandibular region lacking a vascular signal. No focal lesion was found in the orthotopic thyroid gland. Consequently, a contrast enhanced computed tomography (CT) scan was ordered which revealed a round, inhomogeneous, cystic, low density mass with calcifications. The lesion was measured approximately 5 × 3.5 cm. It was located anteriorly to the internal jugular vein and the common carotid artery and it was found to be invasive into the submandibular gland (Fig. 1). There was no relation of the mass with the right lobe of the thyroid. Few regional lymph nodes were enlarged. The thyroid

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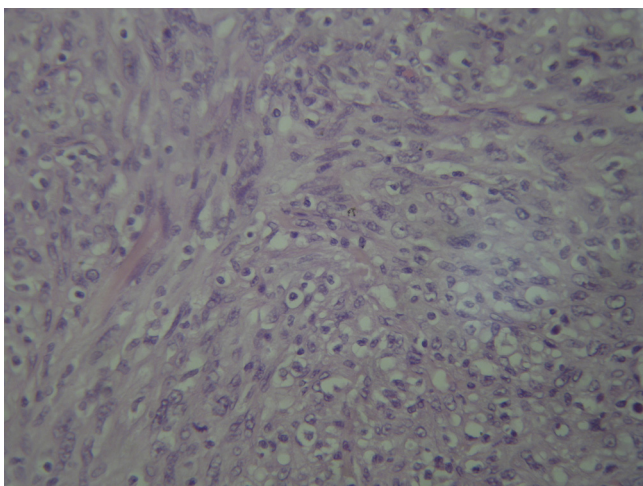
E-mail address: [petkoltsid@yahoo.gr](mailto:petkoltsid@yahoo.gr) (P. Koltsidopoulos).



**Fig. 1.** Computed tomography showing a round, inhomogeneous, cystic, low density mass with calcifications, located anteriorly to the internal jugular vein and the common carotid artery.

gland was normal. Subsequently, fine needle aspiration cytology (FNAC) of the mass was performed and revealed the malignant nature of the lesion. However, no definite conclusion could be made regarding the exact type of carcinoma.

The patient underwent surgery under general anesthesia. The mass was excised along with a wide rim of surrounding tissues. Intraoperative findings revealed that there was no glandular continuity between the thyroid gland and the mass. The postoperative period was uneventful. Histopathological examination demonstrated an anaplastic carcinoma arising within a submandibular lymph node (Fig. 2). Moreover, a focus of normal thyroid tissue was found within another lymph node that was adjacent to the lesion (Fig. 3). In order to determine the extent of the disease, a CT scan of the brain, chest and abdomen was carried out. Metastases were detected in the lungs. The patient was further referred



**Fig. 2.** Tumor shows atypical giant and large cells, spindle cells, mitotic cells and osteoclast-like giant cells. Hematoxylin and eosin stain 440 $\times$ .

to the Oncology Department in order to undergo chemotherapy. The patient died 10 months following diagnosis.

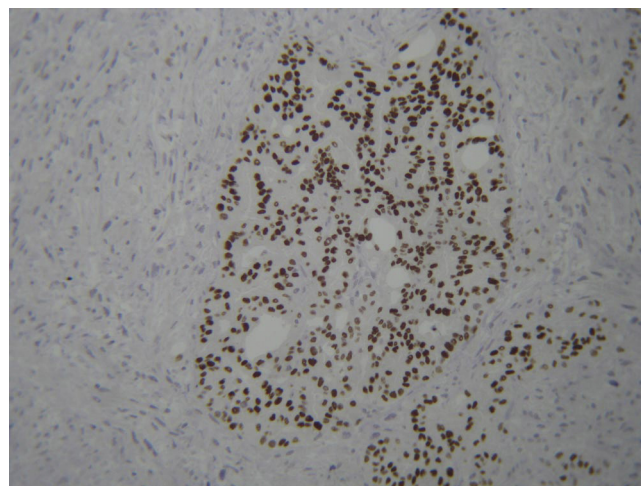
### 3. Discussion

Ectopic thyroid is the most common abnormality of thyroid embryological development and is mainly found in the middle cervical line along the course of thyroglossal duct. Less frequently it can also present in the lateral cervical region, mediastinum, tracheal or oesophageal wall [6]. Ectopic thyroid is susceptible to the same diseases that affect the thyroid gland. Carcinomatous change at an extrathyroid area is a rather uncommon event. The majority of these tumors are papillary. Anaplastic carcinoma arising in ectopic thyroid tissue is extremely rare.

Thyroid carcinoma metastasis should be taken into account in the differential diagnosis of thyroid tissue in the lateral cervical region. Even in case of a thyroid gland without any detectable carcinomatous focus, a micro-carcinoma may be present giving rise to local metastases [7]. Notably, accumulating clinical, pathological, and molecular evidence over the last decades has supported the concept that anaplastic carcinoma may arise from pre-existing differentiated thyroid cancer [8]. This process of “dedifferentiation” may take place either in the primary carcinoma or in a metastasis [9]. There are reports of cases that had primary thyroid papillary carcinoma and anaplastic transformation in metastatic cervical lymph node [10].

In the present case we did not perform a total thyroidectomy considering that FNA could not specify the origin of the tumor. Thus, we could not determine microscopically the presence or absence of a thyroid microcarcinoma. Nevertheless, the option of an ectopic thyroid carcinoma seemed to be higher. Our assumption was mainly based on the fact that a focus of normal thyroid tissue was found within another neighboring cervical lymph node. Moreover, the excised lesion consisted of anaplastic carcinoma lacking follicular or papillary carcinoma. In case of anaplastic transformation from a differentiated carcinoma, there is an intermingling pattern of two neoplastic cell types in the tumor. Therefore, the tumor origin in the present case may be consistent with ectopic thyroid carcinoma.

Furthermore, ectopic thyroid in a branchial cyst should also be taken into account in the differential diagnosis due to the location of the mass. Namely, branchial cysts derived from the first branchial pouch appear in a pre-auricular area in the upper neck, those from the second branchial pouch present as an anterior border tumor



**Fig. 3.** A focus of normal thyroid tissue was found within a cervical lymph node (TTF-1 staining).

of the sternocleidomastoid muscle in the midneck, and those from the third and fourth branchial pouches develop as a supraclavicular tumor in the lower neck.

Moreover, FNA is a significant diagnostic tool contributing to the accurate preoperative assessment of ectopic thyroid tissue, particularly if malignancy is suspected. This examination has high diagnostic accuracy facilitating the appropriate therapeutic approach. Despite that, up to 15% of the samples are nondiagnostic [11]. This limitation is overcome when the biopsy is performed under ultrasound guidance, as compared with biopsy performed using palpation alone. In the present case FNA was not accurately diagnostic. It is important to be aware of the sensitivity and specificity of FNA on one's own hospital.

#### 4. Conclusion

In conclusion, our case highlights the diagnostic challenges that are posed by the presence of anaplastic carcinoma in the submandibular region without any evidence of primary tumor at the orthotopic thyroid gland. In those cases a diagnostic dilemma presents: is it an ectopic thyroid carcinoma or rather a case of occult differentiated thyroid carcinoma metastasis that transformed to anaplastic carcinoma? The answer to this question should be based on the histopathological findings.

#### Conflict of interest statement

All the authors declare no conflict of interest.

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