

CASE REPORT

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Submandibular ectopic thyroid gland: an uncommon presentation

Inès Riahi^{1*} , Rim Fradi¹, Ibtissem Ben Nacef² and Ahlem Blel³

Abstract

Background: Ectopic thyroid is a developmental anomaly of the thyroid gland of embryological origin. Instead of having a pretracheal situation, thyroid tissue is elsewhere, most commonly in the median cervical line along the course of the thyroglossal duct. Lingual thyroid is the most common presentation. Ectopic thyroid tissue in the submandibular region has been rarely reported.

Case presentation: We report herein a case of a 65-year-old man admitted to our department with a complaint of a painless swelling in the left submandibular region.

Conclusions: Thyroid gland ectopia should be considered among the differential diagnoses of submandibular swelling. Ectopic thyroid tissue can present with the same pathology affecting the normal thyroid gland such as malignancy and hyperthyroidism.

Keywords: Thyroid gland, Ectopia, Submandibular region

Background

The thyroid gland develops and descends to its final position, anterolaterally to second and fourth tracheal cartilages, by the eighth week of gestation. A defect of migration can give rise to ectopic thyroid tissue. It is typically encountered in the midline cervical region. Laterally located ectopic thyroid tissue, with or without pathology, is a rare condition. Having a functional orthotopic thyroid gland and submandibular ectopic thyroid tissue is an exceptional event [1].

Case presentation

A 65-year-old man was admitted to our Department of Otorhinolaryngology-Head and Neck Surgery for the management of a painless left submandibular swelling evolving for 4 months. There were no signs of thyroid dysfunction and no history of dysphagia or dyspnea. Physical examination revealed a firm mobile mass of about 4 cm in the left submandibular region with no

other cervical tumefactions besides enlargement of the thyroid gland. Thyroid function tests were normal. Ultrasound of the neck revealed a large echogenic solid lesion of the left submandibular region distinct from the submandibular gland that was normal-looking and a diffusely enlarged heterogeneous thyroid gland suggestive of a goiter. Fine needle aspiration cytology (FNAC) of the submandibular mass was non-diagnostic.

Head and neck computed tomography (CT) revealed a well-defined intensely enhancing soft tissue mass in the left submandibular region (Fig. 1), responsible for a discrete mass effect on the parapharyngeal space medially. The thyroid gland was slightly enlarged with no suspicious nodules or associated cervical lymphadenopathy (Fig. 2).

There was no operative indication for the thyroid gland on the preoperative data. However, a verification of the submandibular mass was required.

The patient underwent a complete surgical resection of the submandibular mass under general anesthesia. The histopathologic finding was normal thyroid tissue and no features of malignancy (Fig. 3).

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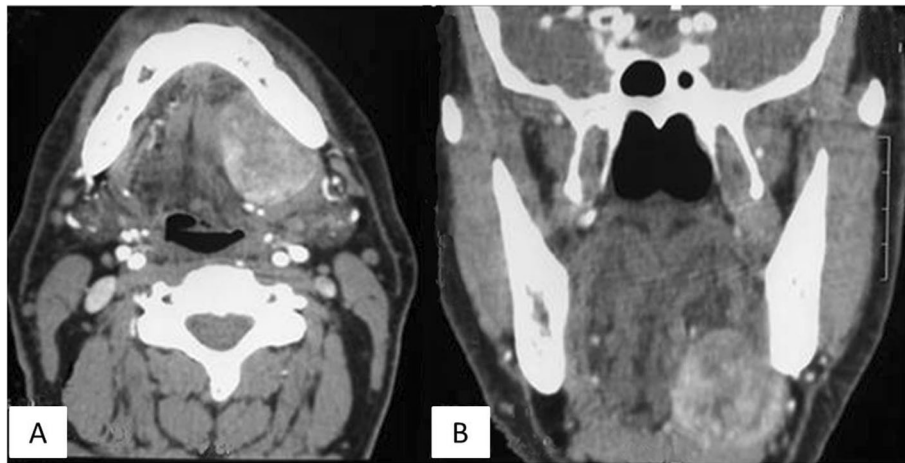


Fig. 1 Post-contrast-computed tomography image. **A** Axial view. **B** Coronal view, showing an enhancing submandibular mass lesion in the left side

The postoperative period was uneventful. The patient was symptom-free and euthyroid over the following year.

Discussion

The thyroid gland is the first endocrine gland to develop in the embryo. It is derived from one median and two lateral anlagen that fuse and entail a middle descent to reach their final position by the eighth week of gestation. Failure of the normal descent of the thyroid gland results in ectopic thyroid tissue [2]. The prevalence of ectopic thyroid tissue is nearly 1/100,000–300,000. The male/female ratio is 1/4. Ectopic thyroid tissue can be seen in any situation on the migration pathway of the thyroid gland, from the foramen caecum to the mediastinum [3, 4]. It is typically encountered in the midline

cervical region. The presence of normal thyroid tissue laterally in the neck has rarely been described [5, 6].

A failure of the lateral anlage to fuse with the median anlage or an aberrant migration with cell rests deposited laterally during the development of the gland can result in the development of lateral aberrant thyroid tissue. Other possible causes include implantation of thyroid tissue during surgery of a normal localized thyroid gland or metastasis of a thyroid carcinoma [7, 8].

The common differential diagnoses of a submandibular mass are lymphadenopathy of various etiologies, submandibular inflammatory or malignant lesions, or tumors of the inferior pole of the parotid gland. Although rare, thyroid ectopia should be considered among the differential diagnoses of a submandibular

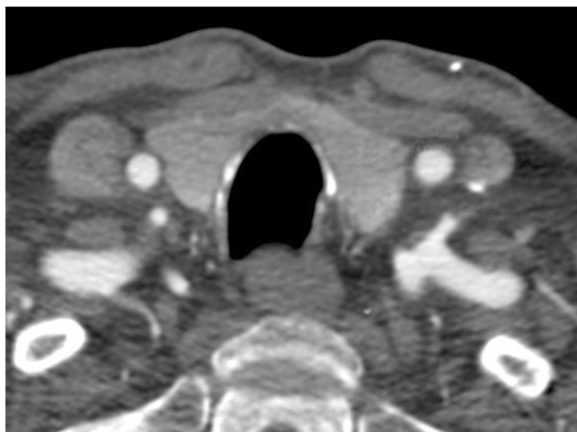


Fig. 2 Post-contrast-computed tomography image. Axial view showing a homogeneously enhancing thyroid gland

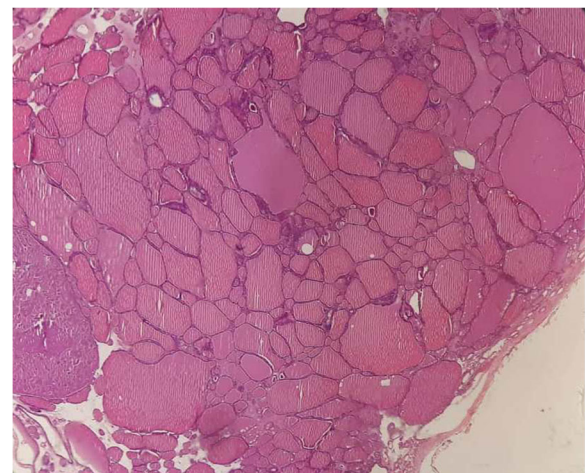


Fig. 3 Thyroid nodule consisting of macro vesicles (× 100)

mass, independent of the submandibular gland, as is the case in our observation [9, 10].

Ectopic thyroids are usually asymptomatic and may become clinically evident with the development of goiters, hyperthyroidism, or malignancy.

Scintigraphy with technetium (Tc 99m) and iodine, in association with ultrasonography and fine needle biopsy (FNB), plays an important role in the diagnosis of ectopic thyroid tissue [11]. Thyroid scintigraphy scanning detects all sites of thyroid tissue as well as hyperfunctional parenchyma [12]. FNB, when conclusive, contributes to the preoperative diagnosis and appropriate therapeutic approach. It has high diagnostic accuracy in the differentiation of benign and malignant processes with an increased sensitivity when biopsy is performed under ultrasound guidance [11].

We managed our case as a submandibular mass of unknown etiology. The likelihood of an ectopic thyroid tissue was low in particular with the evidence of an orthotopic thyroid gland on radiological evaluation. Therefore, scintigraphy was not performed. FNB was non-diagnostic so that surgery became essential.

Submandibular ectopic thyroid has to be differentiated from metastatic thyroid cancer. The rate of malignant transformation in ectopic thyroid is comparable to that in normally located thyroid [13].

Ectopic parenchyma may also be dysfunctional. Eli et al. [14] reported a rare case of ectopic submandibular thyroid causing hyperthyroidism in a patient with a submandibular mass and pre-existing thyroid disease that presents with a deteriorating thyroid function of no apparent reason. The resection of the submandibular mass, which proved to be of a thyroid nature, allowed the correction of the thyroid function.

The therapy of choice for ectopic thyroid tissue is its surgical removal. However, before surgery, it is necessary to ensure that other orthotopic thyroid tissues are functional to avoid the risk of iatrogenic hypothyroidism. In fact, in cases of lateral cervical ectopic thyroid with simultaneous eutopic thyroid, which is a rare situation, the ectopic tissue may be the only functional up to 70% [15, 16].

The treatment of ectopic thyroid depends on its size, local symptoms, functional status of the thyroid gland, and complications [15]. The surgical indications for an ectopic thyroid tissue include the risk of malignancy, refractory hyperthyroidism, signs of compression, or esthetic deformity [17].

Conclusion

Our case highlights the diagnostic and therapeutic challenges that are posed by the presence of ectopic thyroid tissue in the submandibular region. The diagnosis of ectopic thyroid, whether normal or pathological, should be

considered in case of uncommon submandibular tumefaction. The diagnosis should be made, at best, preoperatively, for a better therapeutic decision.

Abbreviations

FNAC: Fine needle aspiration cytology; CT scan: Computed tomography scan; FNB: Fine needle biopsy; Tc 99m: Scintigraphy with technetium 99

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"Not applicable" for this section.

Authors' contributions

IR conceived and designed the manuscript content and supervised the manuscript preparation, editing, and review. RF analyzed and interpreted the patient data regarding the cervical disease and was a major contributor in writing the manuscript. IBN contributed to the literature search and manuscript review. AB supervised and validated the histological data relating part. All authors read and approved the final manuscript.

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Availability of data and materials

The datasets used and analyzed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

Our institution does not require ethical approval for reporting individual cases or case series.

Verbal informed consent was obtained from the patient for the scientific use of medical data.

Consent for publication

Written informed consent was obtained from the patient for its anonymized information to be published in this article.

Competing interests

The authors declare that they have no competing interests.

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