

Case Report

Submandibular ectopic thyroid tissue diagnosed by ultrasound-guided fine needle biopsy

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Abstract: Ectopic thyroid gland is rarely seen in the submandibular region, thereby posing difficult diagnostic and management problems. Two Caucasian women presented with painful swelling in the submandibular region, which increased in size considerably during the preceding months. Ultrasound-guided fine needle biopsy (Ug-FNB) revealed ectopic thyroid tissue. In conclusion, ectopic thyroid gland with or without pathology should be included in the differential diagnosis of submandibular swelling and Ug-FNB should be one of the first diagnostic tools utilized for this rare clinical entity. (J Oral Sci 53, 249-252, 2011)

Keywords: thyroid; ectopia; submandibular region; ultrasonography; biopsy.

Introduction

Thyroid tissue is rarely detected in foreign anatomic locations. Ectopic thyroid tissue is typically encountered in the midline cervical region, but laterally located ectopic thyroid tissue, with or without a normally located thyroid gland, is a very rare condition (1-3). In such cases, the

clinical presentation varies depending on the location of ectopic thyroid tissue (4,5). Here, we present two cases of ectopic thyroid tissue, masquerading as a submandibular swelling, diagnosed by ultrasound-guided fine needle biopsy (Ug-FNB) (Fig. 1). Ug-FNB can contribute significantly to the accurate preoperative assessment of ectopic thyroid tissue occurring in the submandibular region, particularly if malignancy is suspected.



Fig. 1 Ultrasound-guided fine needle biopsy (Ug-FNB). The procedure is performed using a freehand technique without aspiration and without using a guide system or an attached syringe under real-time ultrasound monitoring (photo shows a general Ug-FNB procedure, and does not depict either of the present cases).

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Case Report

Case 1

A 37-year-old Caucasian female presented with pain while swallowing and left palpable submandibular swelling of significant size, which had increased during the preceding months, and was referred for ultrasound examination of the submandibular area and the neck in February 2008. The patient, according to her history, had undergone total thyroidectomy for multinodular goiter 2 years ago and was receiving replacement therapy. According to the latest audit, her thyroidal hormonal check up (T3, T4, T3 resin uptake, TSH) showed normal values. Computed tomography (CT) revealed an inhomogeneous mass on the inside of the left submandibular gland (Fig. 2). Gray-scale ultrasound examination showed that in the left anterior cervical triangle, below the ipsilateral submandibular gland, there was a solid lesion of heterogeneous internal structure and a maximum diameter of 2.2 cm (Fig. 3). Color Doppler ultrasound revealed a small degree of vascularity in the periphery of the lesion. Imaging findings suggested an enlarged lymph node, possibly malignant. The left submandibular gland was normal. In the area of the thyroid gland no residual thyroid tissue was observed. No enlarged lymph nodes were noted in other areas of the neck. Sonography was followed by ultrasound guided fine needle

biopsy, with samples taken from multiple lesion sites, and cytological examination, after staining the samples with a quick Giemsa stain, revealed ectopic thyroid tissue (Fig. 4). The patient underwent surgical ablation of the submandibular swelling shortly afterwards, and histological examination confirmed a diagnosis of residual thyroid tissue in an ectopic position, with no evidence of malignancy. To date, the patient has not experienced symptoms related to the earlier submandibular swelling.

Case 2

A 30-year-old Caucasian female with a painless swelling in the right submandibular region was referred for ultrasound examination of the neck and, if necessary, for Ug-FNB, in November 2008. The submandibular swelling was palpable but invisible and the first diagnostic approach was performed by ultrasound examination. Gray-scale ultrasound examination showed a solid lesion of homogeneous internal structure and a maximum diameter of 2 cm in the right submandibular region. Color Doppler ultrasound showed no vascularity in the lesion (Fig. 5). No swollen cervical lymph nodes or other pathological findings were observed. The right submandibular and thyroid glands were of normal size and morphology. Ug-FNB and cytological examination after quick Giemsa staining confirmed the final diagnosis of ectopic thyroid tissue

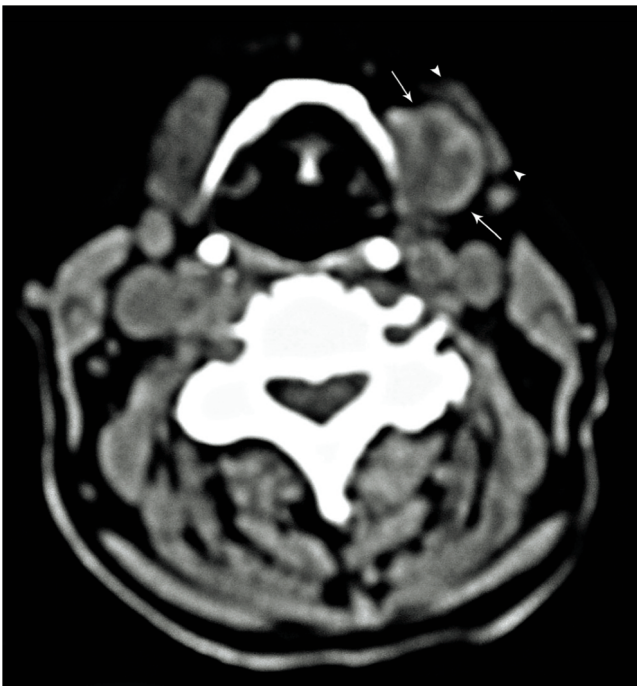


Fig. 2 Case 1: CT showed an inhomogeneous, possibly malignant, soft tissue mass (arrows) on the inside of the left submandibular gland (arrowheads), probably due to an enlarged lymph node.

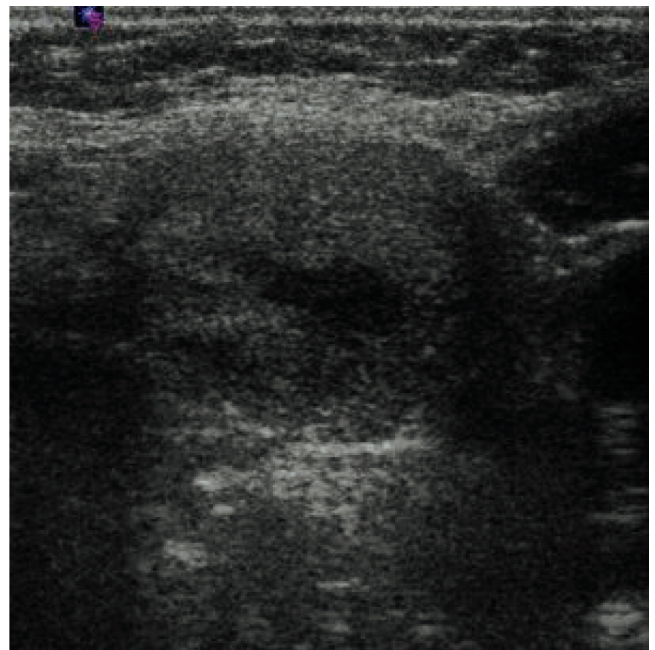


Fig. 3 Case 1: Transverse gray-scale ultrasonogram of the neck: submandibular mass. The mass appears as a submandibular solid lesion with heterogeneous internal structure at the left anterior cervical triangle.

(Fig. 6). The patient underwent surgery based on the Ug-FNB and cytological report. The right submandibular

swelling was removed surgically a week later and histological examination confirmed the cytological diagnosis of ectopic thyroid tissue, without malignancy. To date, the patient has shown no pathology associated with the removed submandibular ectopic thyroid tissue.

Discussion

The thyroid is an endocrine gland located in the lower anterior cervical region, below the thyroid cartilage and in front of the trachea at the level of the 5th, 6th and 7th cervical vertebrae. In rare cases, thyroid tissue may be present in other locations, mainly in the middle cervical line (in the tongue region, within the tongue, under the tongue, near the thyroglossal duct, appearing as a cyst in the thyroglossal duct in front of the larynx, in the trachea), retrosternally (mediastinum, heart) and rarely in the lateral cervical and the submandibular region (2). In rare cases in which the ectopic thyroid tissue is located in the submandibular region, three possible causes have been reported: abnormalities in the formation of the thyroid gland during fetal life, migration of thyroid tissue during surgery in the thyroid gland and metastasis from highly differentiated carcinoma of the thyroid gland (3,6).

Fine needle biopsy with palpation is an important diagnostic tool in the management of patients who present

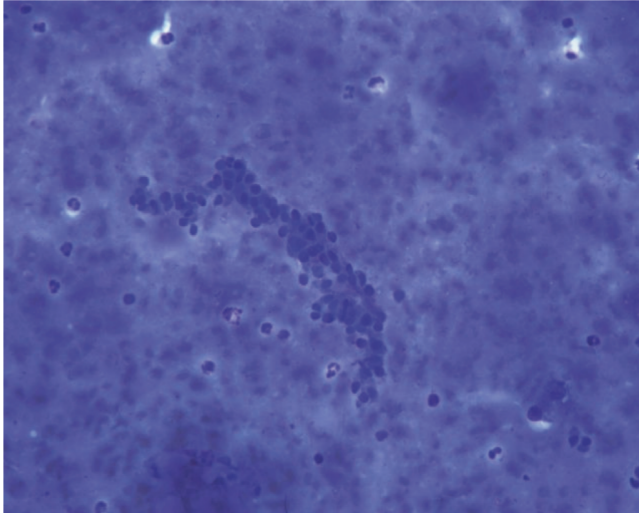


Fig. 4 Case 1: cytological examination (quick Giemsa stain): ectopic thyroid. There are elements of normal thyroid tissue, mostly sparse or dense colloid and a monomorphic population of benign follicular thyroid cell clusters. Follicular cells are cuboidal and slightly larger than lymphocytes, with smooth oval nuclei. The cytoplasm is delicate and naked follicular cell nuclei are often seen on the substrate.

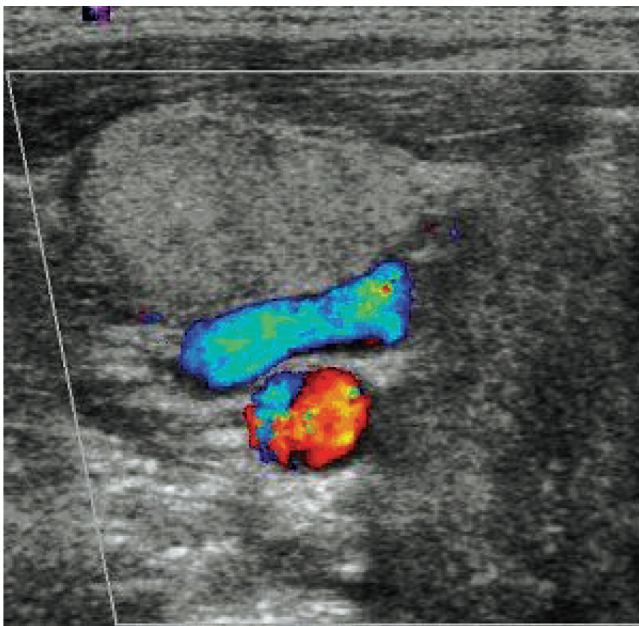


Fig. 5 Case 2: Transverse color Doppler image: submandibular mass at the right anterior cervical triangle. The mass appears as a solid lesion with homogeneous internal structure without vascularity.

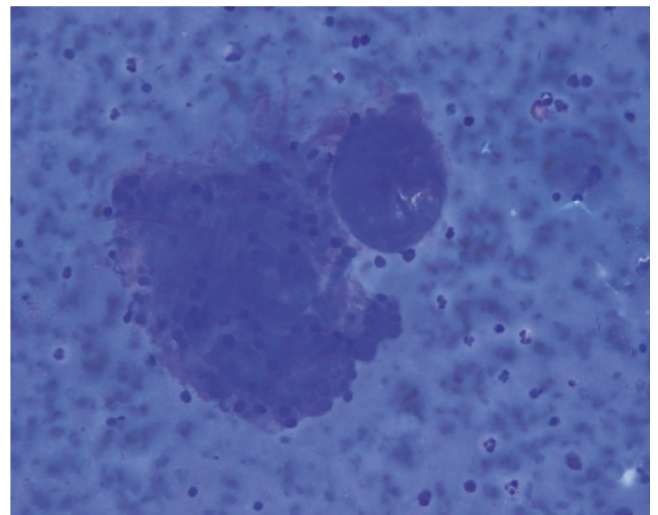


Fig. 6 Case 2: Cytological examination (quick Giemsa stain): ectopic thyroid. There are elements of normal thyroid tissue, mostly sparse or dense colloid and a monomorphic population composed of benign follicular thyroid cell clusters. Follicular cells are often arranged in cohesive flat sheets and follicles. The colloid stains blue or purple with Giemsa staining, and when dense, displays a distinctive cracking artifact resembling a mosaic pattern.

with a submandibular lesion. This examination has high diagnostic accuracy regarding the differential diagnosis of benign and malignant processes in the submandibular region. Despite the effectiveness and diagnostic accuracy of the procedure, up to 15% of the samples are non-diagnostic (7). The problem of non-diagnostic specimens appears to diminish when the biopsy is performed under ultrasound guidance, as compared with biopsy performed using palpation alone, because the number of false-negative samples is reduced and the sensitivity of the method is increased. In short, Ug-FNB contributes significantly to the accurate preoperative assessment of ectopic thyroid tissue occurring in the submandibular region, particularly if malignancy is suspected (8).

Scintigraphy with Technetium (Tc 99m) and Iodine (^{131}I), in conjunction with the above diagnostic methods, plays an important role in the diagnosis and further management and monitoring of patients with ectopic thyroid tissue. The radioisotopic test may highlight both the location of ectopic thyroid tissue and its functionality, and can also raise the suspicion of malignancy. At the same time, the thyroid gland, if present in its normal anatomical position, can be depicted, and its functionality can be assessed. It may also reveal the existence of a second outbreak of ectopic thyroid tissue, which is a critical piece of information for the therapeutic management of these patients (9). The treatment of this pathological entity depends on the clinical presentation, laboratory findings and hormonal profile of the patient, the functionality of ectopic thyroid tissue, the presence and function of the thyroid gland in the normal anatomical location and the suspicion of malignancy (8,10).

In conclusion, our cases of submandibular ectopic thyroid tissue masquerading as submandibular swelling highlight the inherent difficulties in the differential diagnosis of submandibular masses. In summary, the algorithm to differentiate swellings in the submandibular region should include ectopic thyroid tissue with or without pathology, even if there is a normally located thyroid gland. A useful diagnostic tool for this rare clinical entity is Ug-FNB and cytologic examination. This diagnostic method contributes to the preoperative diagnosis and appropriate therapeutic approach, particularly when the ectopic thyroid tissue is

the only functional thyroid gland or when malignancy is suspected.

References

1. Kousta E, Konstantinidis K, Michalakis C, Theodoropoulos GE, Voriias M, Georgiou M, Sambalis G (2005) Ectopic thyroid tissue in the lower neck with a coexisting normally located multinodular goiter and brief literature review. *Hormones (Athens)* 4, 231-234.
2. Nasiru Akanmu I, Mobolaji Adewale O (2009) Lateral cervical ectopic thyroid masses with eutopic multinodular goiter: an unusual presentation. *Hormones (Athens)* 8, 150-153.
3. Babazade F, Mortazavi H, Jalalian H, Shahvali E (2009) Thyroid tissue as a submandibular mass: a case report. *J Oral Sci* 51, 655-657.
4. Kumar R, Sharma S, Marwah A, Moorthy D, Dhanwal D, Malhotra A (2001) Ectopic goiter masquerading as submandibular gland swelling: a case report and review of the literature. *Clin Nucl Med* 26, 306-309.
5. Aguirre A, de la Piedra M, Ruiz R, Portilla J (1991) Ectopic thyroid tissue in the submandibular region. *Oral Surg Oral Med Oral Pathol* 71, 73-76.
6. Feller KU, Mavros A, Gaertner HJ (2000) Ectopic submandibular thyroid tissue with a coexisting active and normally located thyroid gland: case report and review of literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 90, 618-623.
7. Stanek J, Busseniers AE (1993) Fine-needle aspiration diagnosis of ectopic thyroid: report of one case. *Diagn Cytopathol* 9, 59-62.
8. Wang CY, Chang TC (1995) Preoperative thyroid ultrasonography and fine-needle aspiration cytology in ectopic thyroid. *Am Surg* 61, 1029-1031.
9. Sood A, Kumar R (2008) The ectopic thyroid gland and the role of nuclear medicine techniques in its diagnosis and management. *Hell J Nucl Med* 11, 168-171.
10. Kozol RA, Geelhoed GW, Flynn SD, Kinder B (1993) Management of ectopic thyroid nodules. *Surgery* 114, 1103-1107.