CASE REPORTS

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Mediastinal ectopic thyroid tissue as diferential diagnostic problem: A case report

Ektopično tkivo štitaste žlezde u medijastinumu kao diferencijalno dijagnostički problem

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Abstract

Introduction. Mediastinal ectopic thyroid tissue (ETT) represents a rare entity. Clinically, it can manifest with thyroid gland dysfunction or with symptoms and signs caused by a compressive effect on the surrounding structures, but in most cases it is an asymptomatic condition and incidental finding. All pathologic processes, including malignancy that can occur in the orthotopic thyroid gland can also develop in the ETT. **Case report.** We presented a case of a 17-year-old female with incidentally found mediastinal ETT. Besides ETT, the patient had an orthotopic thyroid gland and was euthyroid. During follow-up, mild compressive symptoms developed. Magnetic resonance imaging examination showed a non-significant increase of the mediastinal mass volume, but due to its morphological changes, a suspicion of another etiology was raised. A discrepancy between the positive technetium-99m pertechnetate and negative 131 iodine radionuclide imaging of the mediastinal mass was highly suspicious for malignancy. Surgery was performed and the pathologist confirmed that it was a colloid goiter in the mediastinal ETT. **Conclusion.** Mediastinal ectopic thyroid tissue should be taken into account in the differential diagnosis of the mediastinal tumor mass. An increase in the size of the mediastinal ETT, development of compressive symptoms or suspected malignant alteration require surgical treatment.

Key words:
congenital abnormalities; diagnosis, differential; histological techniques; mediastinal neoplasms; thyroid gland.

Apstrakt

Uvod. Ektopično tireoidno tkivo (ETT) u medijastinumu predstavlja redak entitet. Klinički se može manifestovati simptomima poremećaja funkcije štitaste žlezde ili znacima kompresije okolnih organa, ali najčešće protiče asimptomatski i otkriva se slučajno. Svi patološki poremećaji, koji mogu nastati u ortotopičnoj štitastoj žlezdi, uključujući i malignitet, mogu nastati i u ETT. **Prikaz bolesnika.** Prikazana je 17-godišnja bolesnica sa slučajno otkrivenim medijastinalnim ETT koja je, pored ETT, imala ortotopičnu tireoideu i bila eutireoidna. Tokom praćenja razvili su se blagi simptomi kompresije. Nalaz magnetne rezonance ukazao je na beznačajno uvećanje medijastinalne mase, ali je diferencijalno dijagnostički posumnjano na drugu etiologiju opisane promene. Protivrečnost između pozitivnog nalaza medijastinalne mase ustanovljenog snimanjem tehnecijum-99m pertehnetatom i negativnog nalaza ustanovljenog snimanjem primenom ^{131}joda ukazivao je na mogući malignitet. Bolesnica je operisana, odstranjena joj je medijastinalna masa i patohistološki je potvrđena koloidna struma u medijastinalnom ETT. **Zaključak.** Ektopično tkivo štitaste žlezde u medijastinumu mora se uzeti u obzir prilikom razmatranja diferencijalne dijagnoze medijastinalnih tumorskih masa. Porast medijastinalnog ETT, pojava simptoma kompresije ili sumnja na malignu alteraciju zahtevaju hirurško lečenje.

Ključne reči:
anomalije; dijagnoza, diferencijalna; histološke tehnike; medijastinum, neoplazme; tireoidna žlezda.

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Introduction

Ectopic thyroid tissue (ETT) is a rare congenital anomaly that develops during the migration of the thyroid angle from the floor of the primitive foregut to its final position on the anterior neck between 2nd and 4th tracheal cartilage rings. The prevalence of the ETT is 1 case per 100,000–300,000 people [1], while autopsy studies show the prevalence of 7%–10% in the population. ETT can coexist with or without a normal localized thyroid gland.

The anatomical locations of the ETT can be various: lingual, sublingual, submandibular, lateral cervical space, carotid space, axillar, endotracheal, mediastinal, pulmonary, cardiac, duodenum, stomach, pancreas, porta hepatis, adrenal glands, ovaries even iris and pituitary gland [1–4]. The most common ectopic location is lingual in about 90% of cases [1, 2].

Clinical presentation of the ETT includes both hyper- and hypo-thyroidism, thyroiditis and symptoms caused by compression effect of the ectopic tissue, but it can also be asymptomatic and therefore an incidental finding.

Beside the tests of the thyroid function, imaging methods such as ultrasound, computed tomography (CT), magnetic resonance imaging (MRI), radionuclide thyroid imaging and biopsy, also have an important role in the diagnostic algorithm of the ETT.

Rare locations, functional and morphologic changes in the ectopic tissue can represent a challenge in differential diagnosis, as in this case.

Case report

We presented a 17-year-old female with an incidental finding of two nodules in the left lobe of the thyroid gland on the neck. Ultrasound examination was performed because of repeated sore throats. Further diagnostic procedures included thyroid gland scintigraphy performed with technetium-99m pertechnetate scintigraphy (Figure 1), fine needle aspiration biopsy (FNAB) of the nodules and computed tomography (CT) of the neck and thorax. Thyroid gland scintigraphy with technetium-99m pertechnetate showed normal radionuclide uptake by the gland and uptake by the ETT in the upper mediastinum on the left side. Soft tissue mass just below the left lobe of the thyroid gland, at the level of the *apertura thoracis superior*, without compressive effect on the trachea was described on the CT scan, suspected to be an ETT. Thyroid function was normal, while the additional findings included two hypodense nodules, one in both thyroid gland lobes, with diameter less than 10 mm. Performed FNAB consisted of a benign follicular nodule.

One year later, the patient was examined by an endocrinologist. Ultrasound of the neck showed small cysts (less than 5 mm) in both thyroid gland lobes while the nodules were the same as on the previous examination. Thyroid function was normal (Table 1). Due to the appearance of intermittent pain and feel of pressure in the lower part of the neck, the patient underwent an MRI scan. The MRI scan showed a well-circumscribed soft tissue paratracheal mass

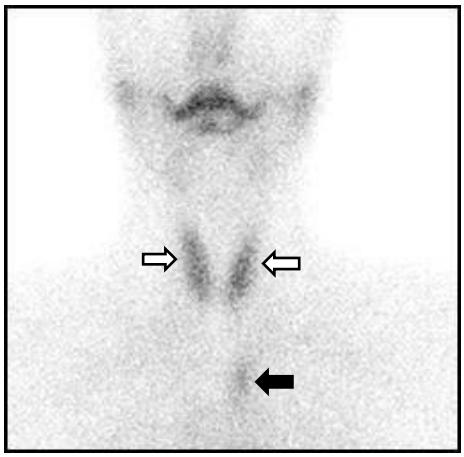

Fig. 1 – Thyroid gland scintigraphy performed with technetium-99m pertechnetate showing normal radionuclide uptake by the gland (white arrows) and uptake by the ectopic thyroid tissue in the upper mediastinum on the left side (black arrow).

Table 1

Results of laboratory examinations before and after surgery

Parameters	Before	After	Reference interval
Free thyroxin (pmol/L)	18.64	17.44	9.0–19.0
Free triiodothyronine (pmol/L)	5.01	5.29	2.6–5.7
Thyroid-stimulating hormone (IU/L)	1.02	1.13	0.35–4.94
Calcitonin (pg/mL)	2.78	n.t.	1.4–78
Anti-TPO antibodues (IU/mL)	< 10.0	n.t.	< 5.6
Anti-Tg antibodies (IU/mL)	< 20.0	n.t.	< 4.1

TPO - thyroid peroxidase; Tg – thyroglobulin; n.t. – not tested.

on the left side (approximately measured 18 × 14 × 30 mm), with inhomogeneous postcontrast enhancement and mild compressive effect on the trachea (Figure 2). Differential diagnosis included ETT, but also teratoma and parathyroid adenoma. Repeated scintigraphy with ^{131}I showed a normal image of the thyroid gland but this time, there was no uptake by the soft tissue mass in the mediastinum (Figure 3). The patient underwent left-sided cervicotomy in general anesthesia, and the surgeon removed the whole mass that was not attached to the thyroid gland. The pathologist confirmed that it was a colloid goiter in the ectopic thyroid gland tissue (Figure 4). After the surgery, thyroid function remained normal (Table 1) and the patient was asymptomatic.

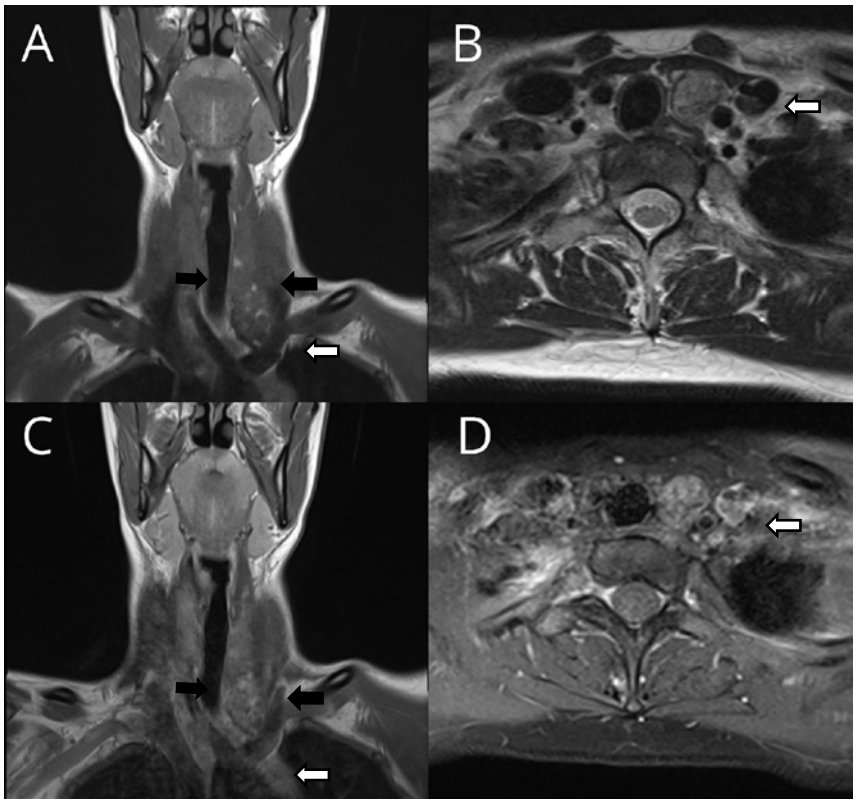

Fig. 2 – Magnetic resonance imaging (MRI) examination: T1 weighted image in the coronal plane (A) and T2 weighted image in the axial plane (B) showing a well-circumscribed paratracheal mediastinal soft tissue mass on the left (white arrows) with mild compressive effect on the trachea; T1 weighted image in the coronal plane (C) and T1 weighted image with fat saturation in the axial plane (D) after gadolinium contrast administration showing an inhomogeneous enhancement of the soft tissue mass (white arrows). Black arrows in A and C indicate normal right and left thyroid gland lobes.

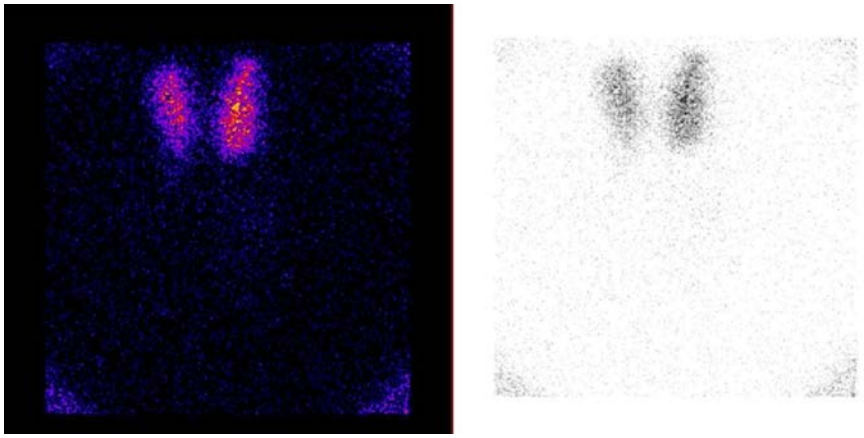

Fig. 3 – Thyroid gland radioiodine scintigraphy with ^{131}I performed with a gamma camera (Symbia E, Siemens) fitted with a high-energy, parallel-hole collimator, 24 hours after oral administration of 1.8 MBq of the radionuclide.

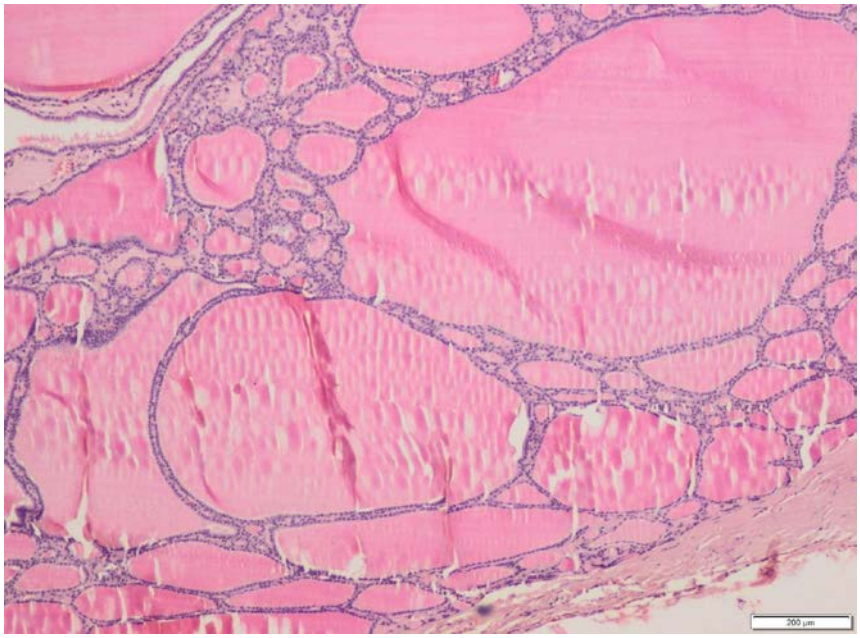

Fig. 4 – Pathohistological finding: Colloid goiter (hematoxylin eosin, × 40).

Discussion

ETT is a rare developmental anomaly. Some studies suggest the genetic base of this anomaly, due to mutation of regulatory genes and transcriptional factors that determine the development of the thyroid gland. Several mutations in genes playing a role during thyroid morphogenesis such as NKX2-1, PAX8, FOXE1, NKX2-5 and TSHR, have been reported, but the molecular mechanisms are not yet fully understood [1, 5, 6]. ETT can appear at any time, but most commonly in childhood, adolescence or in menopause. The female to male ratio is about 4:1 [5]. The presence of a normal thyroid gland in patients with ETT is not necessary. All pathological processes that can develop in the normal gland can also develop in the ETT. Clinical symptoms are typically related to the size and location as well as thyroid function. However, it is mostly an asymptomatic and incidental finding. An increase in the size of the ETT typically correlates with physiological conditions with increased demands for thyroid hormones that is seen during puberty and pregnancy [1, 2].

Mediastinal ETT is an extremely rare entity. To our knowledge only a few cases were reported [5]. It represents about 1% of mediastinal tumors; because of that, it is necessary to be included in the differential diagnosis of mediastinal masses with lymphomas, thymic tumors and dermoid cysts. Hodgkin lymphoma, large B cell lymphoma and lymphoblastic lymphoma are the most common mediastinal lymphomas, while thymic and neuroendocrine carcinomas are rare but highly malignant [7, 8]. Substernal thyroid goiter needs to be differentiated from the ETT. CT and MRI both have a very important role in the diagnosis of ETT, especially when it is distant from the descending pathway of the thyroid [5, 7, 9, 10]. Other imaging modalities such as single-photon emission computed tomography (SPECT CT) with ^{131}I SPECT CT and endoscopic bronchial ultrasound guided biopsy are useful especially in cases of mediastinal ETT [11–14]. Mediastinal ETT can coexist with the orthotopic thyroid gland and in most cases patients are euthyroid, as in our case. If there is no significant mass effect on the surrounding structures and thyroid function is normal, the patients should be followed. In other cases, treatment is surgery. Even in elderly patients, surgical treatment is suggested because of its low risk [7–9]. Both benign and malignant alterations can occur in ETT of any location. Malignancy may occur within ETT with a variety of cell types (papillary, follicular, medullary thyroid cancer, and also Hurtle cell tumor). There are few cases of teratoma and B cell lymphoma in mediastinal ETT [1, 5, 6].

Rarely, a patient with normal TSH can have differences in radionuclide thyroid imaging using technetium pertechnetate vs. iodine scan [15]. This false negative iodine scan could be explained by the presence of the non-organifying thyroid tissue in the ETT (follicular cells which have access to the iodine pump, but without organification).

In our case, the first scintigraphy, at the time of diagnosis, was performed with technetium-99m pertechnetate and the uptake by the ETT was present. The second scintigraphy performed a year later with more sensitive ^{131}I did not show any uptake of the radionuclide. Having in mind the MRI finding, heterogeneous morphology of the tissue, and possible alteration, surgical removal was performed and the pathologist confirmed that it was a benign lesion.

Conclusion

Although mediastinal ETT is rare, it is necessary to be kept it in mind in cases of mediastinal tumor masses. Beside scintigraphy and ultrasound, both CT and MRI have important role in the diagnostic algorithm of ETT. Benign and malignant alterations can occur in ETT of any location. Treatment of mediastinal ETT is either follow-up or surgery, depending on size, location, growth and morphologic changes.

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