

CLINICAL CASE

Adv Clin Exp Med 2004, 13, 4, 725–728
ISSN 1230-025X

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Ectopic Localization of the Thyroid Gland in the Tongue

Nietypowa lokalizacja tarczycy na języku

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Abstract

The authors present a 18-years-old woman with a hormonally active thyroid tissue tumour in the base of the tongue, found accidentally during the laryngological investigation of acute laryngitis. Considering possible complications: dysphonia, dysphagia, dyspnoea, problems with breathing, or even haemorrhage the authors undertook a decision to excise the lingual thyroid and reimplant the tissue fragments under the rectus sheath and in the forearm. Following surgery the patient has not needed hormonal substitution. After 5 years the authors observed subclinical hypothyroidism with thyroid activity present in a small amount of tissue left at the base of tongue and one of the places of the reimplantation (*Adv Clin Exp Med* 2004, 13, 4, 725–728).

Key words: thyroid gland, tongue, reimplantation.

Streszczenie

W pracy przedstawiono przypadek nietypowej lokalizacji tkanki tarczycy u podstawy języka. Nieprawidłowość wykryto przypadkowo podczas badania laryngologicznego ostrego zapalenia krtani u osiemnastoletniej kobiety. Przy rozważeniu możliwych powikłań (dysfonii, dysfagii, duszności, problemów z oddychaniem lub nawet krwotoku) podjęto decyzję o wycięciu tarczycy i ponownym wszczepieniu tkanki poniżej otoczki mięśnia prostego i przedramienia. Po zabiegu chirurgicznym pacjentka nie wymagała zastępczej terapii hormonalnej. Po pięciu latach zaobserwowano subkliniczną niedoczynność tarczycy i aktywność tarczową w niewielkim obszarze tkanki pozostawionej u podstawy języka oraz w jednym z miejsc wszczepienia tkanki (*Adv Clin Exp Med* 2004, 13, 4, 725–728).

Słowa kluczowe: tarczyca, język, replantacja.

Lingual ectopy is the most often pathophysiological localization of the thyroid gland [1]. The thyroid tissue is usually found in the base of the tongue, near the midline foramen caecum, the opening of thyroid-lingual duct in posterior 1/3 of the tongue [2, 3]. Lingual thyroid gland may present with impaired phonation and swallowing, or cause dyspnoea or haemorrhage [4, 5]. In the paediatric population, it is most often found while investigating the cause of congenital hypothyroidism in cases with lower labels of thyroid hormones considering TSH (using in screening test in newborn children) has a highest diagnostic sensi-

tivity and specificity for detection of thyroid dysfunction [6].

In most cases, ectopic thyroid tissue is the only source of thyroid hormones [4, 5, 7].

Both magnetic resonance (MRI) [8], and ultrasonography [9], have been used in searching for ectopic thyroid. Radiolabelled iodine scintigraphy has proved to be the most sensitive test, especially in cases of tumours of little size, not detected during laryngological inspection [10, 11]. Any thyroid tissue in the tongue should be removed. Resection of the ectopic thyroid is indicated even in asymptomatic cases, as it may turn malignant.

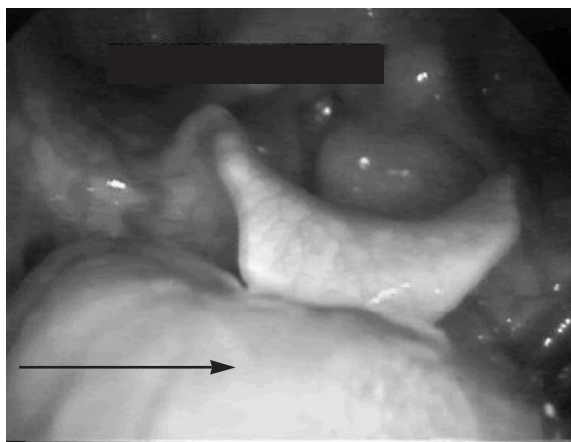
The authors present the case of a surgical removal of the lingual thyroid gland. No hormonal substitution was needed even though not all of the transplanted fragments have engrafted.

Case Report

An 18-years-old woman was admitted to the Department of Endocrinology and Diabetology of Wrocław Medical University for the pre-op evaluation of the lingual thyroid gland. Presence of a lingual mass was incidentally found during the laryngological examination in an outside hospital. Biopsy confirmed the presence of thyroid tissue in the mass. Additional tests showed the TSH level of 3.2 mU/l, normal TRH stimulation test and no thyroid tissue in the anterior neck by ultrasonography. Radiolabelled iodine scintigraphy revealed signs of uptake only in the oral cavity. Due to the risk of malignant transformation, removal of the lingual thyroid tissue was recommended.

Past medical history included childhood asthma that was well controlled and rubeola and measles (with no complications). Laryngological examination revealed a deviation of nasal septum and the 2 cm tumour at the base of the tongue (Fig. 1 a, b).

Tracheostomy with a #7 tube was performed as the first stage of the surgical removal of the tumour. In the second stage, the thyroid gland tissue located at the base of the tongue between foramen caecum and lingual surface of epiglottis was extirpated through the oral approach. Thyroid tissue was liberated with scissors through the longitudinal incision, followed by wedge resection in the direction of epiglottis. The tumour was then excised. The tissue was quite hard and it could not be removed with the capsule. On intraoperative pathology examination, no malignant tissue was found



a

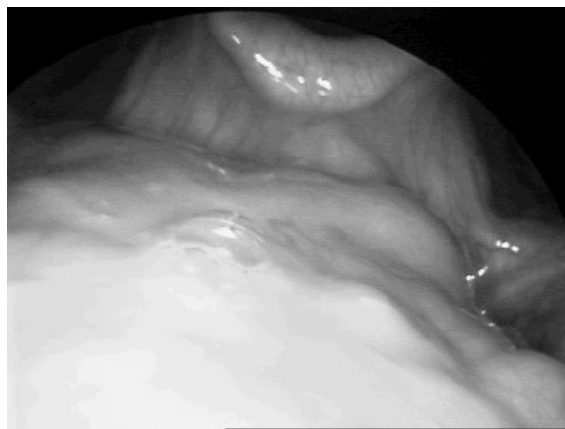


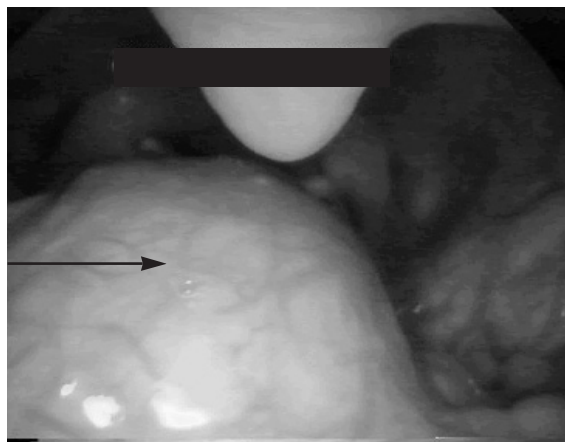
Fig. 2. The base of the tongue after the operation

Ryc. 2. Podstawa języka po operacji

in the specimen. Several fragments of the thyroid tissue were transplanted into the prepared places: under muscle fascia of the left forearm and under rectal sheath of the abdomen. The wound was then closed in layered sutures and dressed. The patient had an uneventful recovery from the surgery (Fig. 2).

Scintigraphy performed 5 years after the surgery showed the presence of active thyroid tissue in the oral cavity at the base of the tongue, left from the sagittal line. There was no significant uptake in the left forearm, and there was some increased uptake at the level of rectal sheath, amounting to twice of the background activity. The total J^{131} uptake after 24 hours was 4%.

Thyrotropin (TSH) and of thyroid hormones (T3 and T4) concentrations were followed after the surgery (Tab. 1). There was a steady increase in free T4 concentration and a steady fall in TSH concentration following the surgery. Throughout the follow-up period, the patient has not required hormonal supplementation.



b

Fig. 1a b. Thyroid gland on the base of the tongue

Ryc. 1a, b. Tarczycza u podstawy języka

Table 1. TSH and thyroid hormones levels following the surgery**Tabela 1.** Stężenie TSH i hormonów tarczycy po operacji

	Directly after the surgery (Bezpośrednio po operacji)	Two years after the surgery (Dwa lata po operacji)	Three years after the surgery (Trzy lata po operacji)
fT3 (pmol/l)	4.65	N/A	4.86
fT4 (pmol/l)	8.17	N/A	11.1
TSH (mU/l)	12.26	6.75	5.75

N/A – not available, fT3 – free T3 concentration, fT4 – free T4 concentration. Normal values: fT3 – 4.0–8.3 pmol/L, fT4 – 9–20 pmol/L, TSH – < 5 mU/l

N/A – brak danych, fT3 – stężenie wolnej trójiodotyroniny T3, fT4 – stężenie wolnej tyroksyny T4. Wartości prawidłowe: fT3 – 4,0–8,3 pmol/l, fT4 – 9–20 pmol/l, TSH – tyreotropina – < 5 mU/l

Discussion

Presented case reminds the clinicians of the possible existence of hormonally active thyroid tissue at the base of the tongue. This tumour location is a result of failure to descend of the embryonic thyroid through the thyreo-lingual duct to its normal position in the anterior neck. This process takes place in the fifth week of intrauterine life, after which the duct regresses [8]. In a large autopsy series, fragments of thyroid tissue of more than 1 cm were found in 10% out of 200 cases; all patients had additional regular thyroid gland in its normal location [9]. Clinically symptomatic lingual thyroid is 7 times more common in women than in men and usually manifests at puberty, menstruation or pregnancy [4, 5, 8]. In described patient, the lingual tumour was found accidentally on otolaryngological examination.

In some cases the lingual thyroid is the only thyroid tissue in the patient [3], most frequently however, it is additional to the regular thyroid gland in the neck [3, 14]. The majority of patients with lingual thyroid are clinically hypothyroid [5, 8]. The patient had subclinical hypothyroidism, and the lingual tumour was the only source of thyroid activity on scintigraphy.

Hypothyroidism in cases of ectopic thyroid may develop later in life [15]. Clinical presentation of a lingual tumour may include dysphonia, dysphagia, dyspnoea, and haemorrhage [1, 2, 4, 5, 16, 17]. Development of these symptoms and the

tendency for malignant transformation both play a role in making decision about surgical removal of the tumour.

Diagnosis of lingual thyroid is made mainly by MRI [10], and, if the tissue is hormonally active, by scintigraphy. The sensitivity of this latter is greater than that of MRI in cases of lingual thyroid. The most frequent modality of treatment is surgical removal of the tumour with reimplantation of the thyroid tissue into the submandibular region or in the lateral wall of the throat.

Both intraoral and transjugular approach with pharyngotomy from subhyoid approach have been used in cases of lingual thyroid [20]. Transjugular approach allows for a complete excision of the ectopic thyroid, especially if it is located deep at the base of the tongue [17]. The ectopic thyroid can also be ablated using radioactive iodine J^{131} [16]. This method is used less frequently as the application of J^{131} may produce transient inflammation and swelling, leading to upper airway obstruction [16].

As the intraoperative histopathologic examination showed no signs of inflammation or malignancy, and as there was no evidence of thyroid gland in the normal location, the patient underwent reimplantation of the excised fragments.

Postoperative scintigraphy showed that the transplanted tissue fragments were not fully hormonally active. Hormonal activity is probably sustained by some amount of tissue left at the base of tongue and by the tissue at the abdominal reimplantation site.

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Received: 16.10.2003

Revised: 6.02.2004

Accepted: 6.02.2004

Praca wpłynęła do Redakcji: 16.10.2003 r.

Po recenzji: 6.02.2004 r.

Zaakceptowano do druku: 6.02.2004 r.