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PRIKAZI SLUČAJEVA

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LINGUAL THYROID IN A YOUNG ASYMPTOMATIC FEMALE PATIENT WITH HYPOTHYROIDISM – CASE REPORT

LINGVALNA ŠTITASTA ŽLEZDA KOD MLADE PACIJENTKINJE BEZ SIMPTOMA SA HIPOTIREOIDIZMOM – PRIKAZ SLUČAJA

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Summary

Introduction. Lingual thyroid is a rare condition where thyroid tissue is abnormally located at the base of the tongue due to embryological development issues. The precise prevalence is uncertain, as many asymptomatic individuals do not undergo clinical examination. **Case report.** We present a case of a 28-year-old woman with a history of hypothyroidism. The patient exhibited no symptoms indicative of lingual thyroid. A neck ultrasound identified hypoplastic thyroid tissue centrally in the neck and a solitary nodule in the submandibular region. Further evaluation with an oropharyngeal examination revealed a solid mass at the base of the tongue. A technetium-99m pertechnetate thyroid scintigraphy, performed with single photon emission computed tomography on a hybrid gamma camera, demonstrated functional thyroid tissue at the tongue's base, measuring 16 mm in diameter, with no functional glandular tissue in the central neck region. **Conclusion.** Asymptomatic lingual thyroid can be diagnosed through oropharyngeal examination, neck ultrasound and technetium-99m pertechnetate thyroid scintigraphy using a hybrid gamma camera, which provides both functional and anatomical data. Management should be individualized based on the patient's symptoms and thyroid hormone levels.

Key words: Lingual Thyroid; Hypothyroidism; Thyroid Dysgenesis; Diagnosis; Ultrasonography; Radionuclide Imaging

Sažetak

Uvod. Lingvalna štitasta žlezda je retka ektopična lokalizacija štitaste žlezde koja se javlja u vidu čvora na bazi jezika zbog abnormalnosti u embriogenezi. Tačna učestalost lingvalne štitaste žlezde nije poznata jer mnogi asimptomatski pojedinci nikada ne dođu na klinički pregled. **Prikaz slučaja.** Prikazan je slučaj pacijentkinje stare 28 godina koja se javila na naše odeljenje sa kliničkom istorijom hipotireoze. Pacijentkinja nije prijavila nikakve simptome koji bi ukazivali na prisustvo lingvalne štitaste žlezde. Ultrasonografskim pregledom vrata otkriveno je hipoplastično tkivo štitaste žlezde u centralnom delu vrata i solitarni čvor u submandibularnoj regiji. Radi dodatne evaluacije urađen je orofaringealni pregled kojim je uočena solidna masa na bazi jezika. U našoj službi izvršena je studija scintigrafije štitaste žlezde tehnecijumom-99m pertehnetatom uz upotrebu jednofotonske emisione kompjuterizovane tomografije na hibridnoj gama kameri. Slike su pokazale funkcionalno tkivo štitaste žlezde u predelu baze jezika sa većim prečnikom od 16 mm i odsustvo funkcionalnog žlezdanog tkiva u centralnoj regiji vrata. **Zaključak.** Prisustvo lingvalne štitaste žlezde kod pacijenata bez simptoma može se dijagnostikovati putem orofaringealnog pregleda, ultrazvučnog pregleda vrata i scintigrafije štitaste žlezde tehnecijumom-99m pertehnetatom uz upotrebu hibridne gama kamere koja istovremeno pruža funkcionalne i anatomske informacije. Lečenje pacijenata sa lingvalnom štitastom žlezdom treba da bude personalizovano i zasnovano na simptomima pacijenta i statusu tiroidnih hormona.

Ključne reči: lingvalna štitasta žlezda; hipotireoidizam; ektopična štitna žlezda; dijagnoza; ultrasonografija; radionuklidni imidžing

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Introduction

Lingual thyroid (LT) is a rare condition characterized by the presence of thyroid tissue at the base of the tongue, resulting from its incomplete migration from the foramen caecum to the pretracheal region during embryogenesis [1]. The occurrence rate of LT ranges from approximately 1 in 100,000 to 300,000 individu-

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Abbreviations

LT	– lingual thyroid
CT	– computed tomography
MRI	– magnetic resonance imaging
FNAC	– fine-needle aspiration cytology
FT4	– free thyroxine
TSH	– thyroid-stimulating hormone
SPECT/CT	– single photon emission computed tomography/ computed tomography
EET	– ectopic thyroid tissue

als, with a higher prevalence among females [2, 3]. Despite these estimates, the exact frequency of LT remains uncertain due to the large number of asymptomatic individuals who do not seek clinical evaluation.

Although most patients with LT are asymptomatic, symptoms can arise from the enlargement of the glandular tissue. Common symptoms include a sensation of a lump in the throat, dysphagia, dysphonia, coughing and loud snoring [1, 4]. Approximately one-third of individuals with ectopic thyroid tissue exhibit clinical manifestations of hypothyroidism due to its typically inadequate production of thyroid hormones by the ectopic tissue [1, 5]. In about 70% of cases, LT is the only functioning thyroid tissue [6], and the simultaneous presence of LT and a normally positioned thyroid gland is extremely rare [7, 8]. The diagnosis of LT typically involves a combination of clinical assessment and various diagnostic procedures, including biochemical tests, neck ultrasonography, thyroid scintigraphy, computed tomography (CT) scans, and magnetic resonance imaging (MRI).

Figure 1. SPECT scan image, lateral view showing intense technetium-99m uptake in the tongue base area
Slika 1. *SPECT sken slike, lateralni snimak – prikazuje intenzivno nakupljanje tehnecijuma-99m u predelu baze jezika*

Figure 2. SPECT/CT fusion images showing intense technetium-99m uptake in the tongue base area
Slika 2. *SPECT/CT fuzione slike pokazuju prikazuje intenzivno nakupljanje tehnecijuma-99m u predelu baze jezika*

Fine-needle aspiration cytology (FNAC) also plays crucial role in confirming the diagnosis [1, 9, 10].

In this case report, we present a rare instance of a young female patient with hypothyroidism who exhibited non-functional thyroid tissue in its typical cervical location alongside functional ectopic LT tissue, despite being asymptomatic.

Case report

We present a case of a 28-year-old female who was referred to our department for further evaluation due to clinical concerns suggesting a possible presence of a lingual thyroid (LT).

Suspicion of LT arose from initial ultrasound findings and an oropharyngeal examination. A neck ultrasound revealed hypoplastic thyroid tissue in the central cervical region and a solitary isoechoic nodule in the submandibular area. Additional evaluation with an oropharyngeal examination revealed a solid mass at the base of the tongue. There was no enlargement of the cervical lymph nodes, and other physical examination findings were unremarkable. The patient's medical history indicated a diagnosis of hypothyroidism at the age of 12, with consistent adherence to levothyroxine replacement therapy since that time. The daily dosage of levothyroxine has consistently been 25 mcg. At the most recent check-up, laboratory tests showed a free thyroxine (FT4) level of 13.04 pg/dL (normal range, 9-19 pg/dL), and a thyroid-stimulating hormone (TSH) level of 1.05 IU/mL (normal range, 0.35-4.94 IU/mL). The patient did not report any symptoms indicative of LT.

In our department, we performed a single photon emission computed tomography/computed tomography (SPECT/CT) scan with technetium-99m pertechnetate. The SPECT and SPECT/CT fusion images revealed functional thyroid tissue at the base of the tongue with a diameter of 16 mm, and no functional glandular tissue in the central cervical position **(Figures 1 and 2)**. This confirmed the diagnosis of LT ectopia. Given the absence of symptoms, we recommended regular follow-up for the patient, including periodic assessment of hormonal levels and ultrasound examinations of the neck.

Discussion

The thyroid gland begins its development around the 24th day of gestation, making it the earliest en-

docrine gland to form in the human body. Occasionally, during embryonic development, the thyroid gland fails to properly migrate from its initial position at the base of the tongue to its final pre-tracheal location in the neck. This anomaly results in the presence of functional thyroid tissue in an atypical location, known as ectopic thyroid tissue (ETT) [2, 11, 12]. EET can be found along the thyroid's descent path, either in the midline or laterally within the neck. It may also occur in more distant regions, such as the mediastinum, beneath the diaphragm, or in various other locations throughout the body [2]. Approximately 90% of the reported ETT cases are found at the base of the tongue [13]. The first documented case of ectopic thyroid in a newborn reported by Hickman in 1869 [7]. The exact factors contributing to the failure of descent in cases of LT are still unclear. However, some researchers suggest a potential link between maternal antithyroid immunoglobulins and impaired gland descent during early fetal development [5]. Additionally, molecular irregularities, including mutations in genes crucial for thyroid development and differentiation, may also play a role in these migration anomalies, although more extensive investigations are necessary. For instance, the expression levels of certain genes vary depending on the anatomical location of the thyroid tissue [14].

Diagnosing LT presents a clinical challenge due to its often asymptomatic nature and atypical presentation [1]. In our case, the diagnosis of LT ectopia was confirmed through a combination of clinical examination and diagnostic imaging, consistent with previous reports [1, 5, 6]. Approximately 33% of patients with LT are hypothyroid and without remarkable clinical manifestations [5], as was the case with our patient. Inadequate blood supply to the ectopic thyroid gland, necessary for normal function, along with iodine organification defects, are potential causes of hypothyroidism [15]. The absence of symptoms in our patient can be attributed to adequate substitution therapy, which generally maintains the thyroid gland's volume [16]. In 93% to 100% of LT cases, the orthotopic thyroid tissue is absent [17]. However, this was not observed in our patient, as the neck ultrasound revealed the presence of a hypoplastic thyroid gland in its typical pre-tracheal location. Despite the presence of thyroid tissue in its orthotopic location, SPECT/CT images revealed its dysfunctionality, with the LT tissue at the base of the tongue being the only functional thyroid tissue. These findings are consistent with previous research, indicating that if LT tissue is present, it is often the only functional thyroid tissue in the body [1, 3, 6]. In our case, FNAC confirmation was not required, but the potential importance of FNAC should be considered, especially if suspicious focal changes in the LT tissue are present, as cases of thyroid carcinoma originating from ETT have been reported [6, 18]. Certain ultrasound features in thyroid nodules, such as marked hypoechogenicity, irregular margins, microcalcifications, and a taller-than-wide shape, are commonly associated with a higher risk of malignancy. Consequently, clinicians should consider performing FNAC in these cases [19].

The management of LT depends on several factors, including the presence of symptoms, the size and location of the ETT, and the patient's thyroid function status. In asymptomatic cases with preserved thyroid function, regular monitoring of function and morphology may be appropriate. In patients with hypothyroidism, hormone replacement therapy and follow-up is considered sufficient if there are no other symptoms, as was the case with our patient. However, symptomatic patients may require intervention, which may include surgical excision or radioiodine therapy [1, 11, 20]. Radioiodine ablation is recommended for those presenting with high-risk symptoms. In cases where symptoms are resistant to conservative therapy, malignancy is suspected, or high-risk symptoms preclude the use of radioiodine, surgical intervention should be considered [20, 21].

Conclusion

Lingual thyroid in asymptomatic patients can be effectively diagnosed through oropharyngeal examination, neck ultrasound, and technetium-99m pertechnetate thyroid scintigraphy using a hybrid gamma camera that provides both functional and anatomical information. The management of patients with lingual thyroid should be personalized, taking into account the patient's symptoms and thyroid hormone status.

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