Abstract:
Ectopic thyroid tissue can be found anywhere between the foramen cecum and the normal position of the thyroid gland. Although very uncommon, it is most often found in the region of the foramen cecum, in patients in whom the gland fails to descend. It may present with symptoms of dysphagia, upper airway obstruction, or even hemorrhage at any time from infancy through adulthood. We present the case of a twelve-year-old male child with a midline cervical mass clinically presented with high dysphagia suffering for one year. Dysphagia worsened over the past two months and was accompanied by increasing in nocturnal dyspnoea and recent onset of sleep apnea. Elements in the diagnostic and therapeutic evaluation are described with attention to the clinical findings, laboratory tests, and radiographic imaging studies employed in confirming the diagnosis and planning appropriate treatment. The natural history of the condition is reviewed and a treatment strategy is outlined that focuses on the use of suppressive doses of thyroid hormone as the initial therapy. Surgical excision of the gland is reserved for more advanced cases of gland enlargement resulting in airway compromise, severe dysphagia that limits oral intake, or ongoing hemorrhage.

Key words: Lingual thyroid.

Introduction:
Lingual thyroid (LT) is a rare developmental disorder due to the aberrant embryogenesis, during the descent of the thyroid gland from the foramen caecum of tongue to its prelaryngeal site in the neck. Ectopic thyroid indicates the presence of the thyroid tissue in other locations than the anterior neck region, which is its normal position. It is a very rare entity.

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Case Report:
A 12-year-old boy without any past medical history consulted for high dysphagia suffering for one year. Dysphagia worsened over the past two months and was accompanied by increasing in nocturnal dyspnoea and recent onset of sleep apnea. There were neither signs of thyroid dysfunction nor alteration of general condition. Its frequency is estimated at 1/4000 to 1/8000 among patients with hypothyroidism and 0.3% of all diseases of the thyroid gland. It particularly affects young women. Its occurrence in children is rare. In 1869, Hickman reported the first case with lingual thyroid on a newborn baby. Diagnosis is mainly based on clinical examination and imaging. Treatment is mainly medical and must take into account the physiological requirements for thyroid hormones. In this study, we report a case of lingual thyroid and we review the literature on this topic.

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Cervical CT (Fig.-2) scan showed a rounded lesion located at the base of the tongue, with heterogeneous enhancing calcification. Thyroid compartment was empty. Thyroid scintigraphy with technetium (Tc99m) showed intense and elective uptake of radiotracer at the area of the base of the tongue and no uptake in the normal thyroid location. Hormonal tests showed hypothyroidism with lower level of FT4 (15.4pmol/L) and increase of TSH (5.2IU/mL). Treatment began with the administration of L-thyroxine for 3 months. The management of the case was discussed fully with the patient and his family & they agreed for surgery. Elective surgical resection of the thyroid gland was performed through a suprathyroid external approach under general anesthesia. A nasotracheal intubation was done before surgery and tracheostomy was done before recovery from anaesthesia. The surgery and postoperative period were uneventful. Decannulation of tracheostomy was started on 3rd postoperative day and tracheostomy closure was done on 5th postoperative day. The patient was discharged on the 5th postoperative day, under L-thyroxine supplement treatment as the endocrinologists recommended. The specimen was sent for histopathology analysis and the result was ectopic thyroid tissue compatible with lingual thyroid.

Discussion:

During embryogenesis, the thyroid tissue descends from the foramen caecum area of the tongue base to the lower part of the neck. This descent process may be impaired anywhere along this path of primitive thyroglossal duct. Maternal antibodies against thyroid antigens are thought to be the cause of the failure in the descending process of thyroid tissue from its origin to its final position. Although it is usually found along the normal path of development it can be found in the mediastinum, heart, oesophagus, porta hepatis, ovary, etc. The incidence is reported to be 1:100000-300000 persons, 7 times higher in females than males.

Ectopic lingual thyroid is commonly detected during periods of increased demand for thyroid hormones, for example adolescence and pregnancy. In our case it has become symptomatic during puberty. Dysphagia, pain, dyspnoea, bleeding and fullness in the throat are the major signs and symptoms. Usually they represent a nodular mass in the base of the tongue. The investigations include thyroid function tests, ultrasonography and scintigraphy. Scintigraphy by using technetium (Tc-99m), I-131, or I-123, is an important diagnostic tool to detect ectopic thyroid tissue and to show the absence or presence of normally positioned thyroid tissue. Fine needle aspiration cytology (FNAC) confirms the diagnosis of ectopic thyroid. It is the only modality to differentiate between benign and malignant lesion. For cases completely asymptomatic and euthyroid regular follow-up is recommended in order to detect mass complications. For mild symptoms and hypothyroidal states, levothyroxine replacement therapy may be effective. Surgical treatment of lingual thyroid depends on size and local symptoms as well as on other parameters such as patient's age, functional thyroid status and complications of the mass. Several surgical approaches have been described such as transoral route, transhyoid route, suprathyroid and lateral pharyngotomy. The transoral route is the preferred approach avoiding deeper structures complications such lingual nerve injury and deep cervical infections.

Conclusion:

Lingual thyroid as a rare developmental anomaly should be included in the differential diagnosis of tongue masses especially in children and adolescence during periods of increased thyroid hormone demand. Investigations include thyroid function tests, neck ultrasound and neck CT. FNAC is not preferred by some authors because of bleeding possibility. Transoral approach provides good exposure, being less traumatic for the patient with better postoperative recovery.
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