



## THYROID TISSUE IN BUCCAL MUCOSA: A RARE PRESENTATION OF THYROID ECTOPIA

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### Abstract:

Ectopic thyroid tissue is a rare entity resulting from developmental defects at early stages of thyroid gland embryogenesis, during its passage from the floor of the primitive foregut to its final pre-tracheal position. It is frequently found along the course of the thyroglossal duct or laterally in the neck, as well as in distant places such as mediastinum and subdiaphragmatic organs. Presence of thyroid tissue in buccal mucosa is a rare presentation of thyroid ectopia. No such case has been reported in the world literature. We report a case of ectopic thyroid tissue in buccal mucosa in a seven years old male child who presented with a gradually increasing swelling in the right buccal mucosa. Aspiration cytology was suggestive of squamous papilloma.

This also protects the recurrent laryngeal nerve from damage. Capsular dissection helps in protecting the recurrent laryngeal nerve and also protects the blood supply of parathyroid glands.

Minimally invasive transoral excision with CO<sub>2</sub> laser was done and the tissue was sent for histopathology which revealed heterotopic thyroid tissue with follicular adenoma. Thyroid function tests suggested euthyroidism. Ultrasonography of the neck showed a normal thyroid gland in its normal location.

## Introduction:

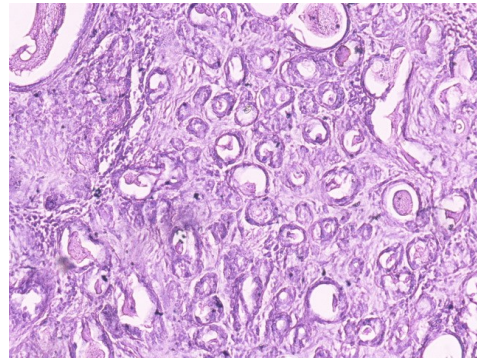
Ectopic thyroid tissue is a rare entity resulting from developmental defects at early stages of thyroid gland embryogenesis, during its passage from the floor of the primitive foregut to its final pre-tracheal position. It is frequently found along the course of the thyroglossal duct or laterally in the neck, as well as in distant places such as mediastinum and subdiaphragmatic organs.[1] Presence of thyroid tissue in buccal mucosa is a rare presentation of thyroid ectopia. No such case has been reported in the world literature. We report a case of ectopic thyroid tissue in buccal mucosa in a seven years old male child who presented with a gradually increasing swelling in the right cheek.

## Case Report:

A seven years old male child presented with the history of a swelling in the right buccal mucosa first noticed about eight months back. The swelling was gradually increasing in size with no history of pain, ulceration or bleeding from the swelling. Birth history and developmental milestones were normal. A detailed general and systemic examination did not reveal any abnormality. Local examination revealed a 1 × 1 centimetre, solid, soft, smooth margined, mobile and non tender mass covered with intact mucosa in the right cheek.

Fine needle aspiration cytology (FNAC) was done which was suggestive of squamous papilloma. The child underwent minimally invasive trans-oral excision of mass using CO2 laser.

The tissue was sent for histopathology which revealed heterotopic thyroid tissue with follicular adenoma (Figure 1). Since the diagnosis of ectopic thyroid was made post-operatively, the child was further evaluated for the presence of eutopic thyroid tissue and thyroid function. Thyroid function tests suggested euthyroidism. Ultrasonography of the neck showed a normally located thyroid gland which was normal in size, shape and echotexture.



HPE picture showing hypertrophied thyroid tissue

## Discussion:

Ectopic thyroid refers to the presence of thyroid tissue in locations other than the normal anterior neck region between the second and fourth tracheal cartilages. It is the most frequent form of thyroid dysgenesis, accounting for 48-61% of the cases.[2]

In 1869, Hickman reported the first case of ectopic thyroid tumour of the base of the tongue, pressing down the epiglottis on the larynx and causing death by suffocation sixteen hours after birth.[3] Prevalence of this condition is reported to be between 1 per 100,000-300,000 persons. To date, about 500 cases have been reported in the literature.[4] Lingual thyroid is the most common type accounting for 90% of cases, while sublingual types which may be suprahyoid, infrahyoid or at the level of the hyoid bone are less frequently encountered.[5] Other locations include the trachea, submandibular, lateral cervical regions, axilla, palatine tonsils, heart, ascending aorta, thymus, oesophagus, adrenal gland, ovary, fallopian tube, uterus and vagina.[6] But the presence of thyroid tissue in the buccal mucosa has not been reported in the literature.

Thyroid gland is the first of the body's endocrine glands to develop, as a proliferation of endodermal epithelial cells on the median surface of the developing pharyngeal gut between the 1st and 2nd pharyngeal pouches and descends in front of the hyoid bone and the laryngeal cartilages to reach the final position in front of the trachea in the 7th week of gestation. Although the molecular mechanisms involved in thyroid dysgenesis are not fully known, genetic research has shown that the gene transcription factors TITF-1(Nkx2-1), Foxe1 (TITF-2) and PAX-8 are essential for thyroid morphogenesis and differentiation. Mutation in these genes may be involved in abnormal migration of the thyroid or heterotopic differentiation of uncommitted endodermal cells.[2,7]

Ectopic thyroid may become goitrous and may be associated with either hypofunction or hyperfunction. Sometimes, benign or malignant neoplastic changes can occur in ectopic thyroid tissue. The majority of these tumours are described as being of the follicular type, while papillary forms comprise 23%. This is in contrast to normal thyroid gland neoplasms, of which papillary tumours form the predominant form.[6]

Radionuclide thyroid imaging employing technetium-99m pertechnetate, iodine-131 or iodine 123 is useful in the evaluation for ectopic thyroid. Thyroid tissue takes up the radioisotope and this helps in localizing the ectopic thyroid and at the same time in determining the presence of a eutopic thyroid gland.[4] High resolution ultrasound scanning is also favoured in the initial assessment, especially in patients presenting with neck masses. It is non-invasive, cost-effective and does not expose patients to ionizing radiation. At the same time it can be used to determine the presence of a eutopic thyroid.[8] CT scan and MRI are useful when a eutopic thyroid gland is not identified by ultrasound. Tissue biopsy for histology or fine needle aspiration cytology (FNAC) is important to confirm the diagnosis and rule out malignancy.[9]

Asymptomatic euthyroid patients with ectopic thyroid do not usually require therapy but are kept under observation.

In patients with hypothyroidism, suppressive therapy is administered using exogenous thyroid hormone. This suppresses the TSH level and causes reduction in the size of the gland. Euthyroid patients with mild obstructive symptoms can also benefit from suppressive therapy.[10]

Surgical intervention is indicated when severe obstructive symptoms, bleeding, ulceration, cystic degeneration and malignancy occur or for cosmetic reasons. It is important to determine the presence of a normally located thyroid gland before removal of the ectopic tissue to avoid hypothyroidism.[4,11] Radioactive iodine 131 therapy is an alternative to surgical ablation. It is indicated in patients who are not fit for surgery, in those who refused operation and where surgical resection is not feasible due to anatomical difficulties. It is contraindicated in pregnant women and avoided in younger paediatric patients.[10]

Ectopic thyroid remains a rare entity with buccal mucosa as a site of presentation reported nowhere in the literature. Therefore we decided to report this case and have a brief review about its various aspects.

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