

Case report – Lymphoma of lingual thyroid

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Abstract

Ectopic thyroid is an abnormal embryological development of unknown pathology. Lingual thyroid is the commonest ectopic thyroid. Symptoms may vary depending on individual, the spectrum of presentation includes mild throat discomfort to upper airway obstruction. Malignant transformation of ectopic thyroid especially to lymphoma is extremely rare. We present a case of 70 year old gentleman with muffled voice, globus sensation, right neck swelling and shortness of breath. Diagnosis was confirmed with Technitium (Tc-99) Scintigraphy as sublingual thyroid. Core biopsy of the neck mass revealed as diffuse large B cell lymphoma. He was then staged and chemoradiotherapy was planned and commenced.

Keywords

Ectopic thyroid; ectopic thyroid lymphoma; malignant transformation; technitium scintigraphy; chemoradiotherapy.

Introduction

Lingual thyroid is a rare embryological anomaly. It is failure in thyroid gland to descend from foramen caecum to its prelaryngeal position, frequently known as the “ectopic” thyroid. The prevalence in the general population is 1 in 100,000 with female preponderance [1]. The commonest site is at base of tongue (90%) while other sites can be in the submandibular gland, cervical lymph node and trachea [1]. The pathogenesis of this anomaly is unclear. Hypothyroidism is typically seen in patients with lingual thyroid [2]. Most patients are asymptomatic but can present with dysphagia, dysphonia, throat discomfort and occasionally respiratory obstruction. Technitium (Tc-99) Scintigraphy is the imaging of choice used to demonstrate ectopic thyroid [2]. Ectopic Thyroid Lymphoma (ETL) is rare. It accounts for 2 cases in 1,000,000 and reported to be less than 5% of all thyroid malignancies [3]. ETL B- cell lymphoma is more prevalent than T- cell lymphoma. In this case study, unexpected ectopic thyroid lymphoma is reviewed.

Case Report

A 70 year old gentleman with no past medical illness presented to ORL clinic with history of muffled voice and fullness in the throat for 3 years, associated with intermittent shortness of breath for 1 month duration. Further history revealed that patient had right neck swelling for 2 weeks which progressively increased in size. Clinical examination of the neck noted swelling at level II and IV, firm and painless. Fiber optic nasopharyngolaryngoscopy showed large cystic mass measuring 5 x 5 cm occupying base of tongue and vallecula (Figure 1). Laryngeal structures were pushed to the left side. Despite having a large mass at the base of the tongue, he was not stridorous but has muffled voice. Aspiration from the cystic mass yielded hemoserous aspirate and was reported as atypical lymphoid cell suggestive of lymphoma. The mass shrunk after aspiration thus relieving the airway symptom. Fine Needle Aspiration Cytology (FNAC) of right neck was done and reported as atypical lymphoid cells. Core biopsy of right neck swelling came to be diffuse large B cell lymphoma. Thyroid function test confirms the patient to be hypothyroid. Contrast Enhanced Computed Tomography (CECT) of the neck reported a large, rim enhancing cystic lesion occupying the vallecula with mass effect to surrounding structures and cervical lymphadenopathy (Figure 2). There was absence of thyroid gland in the expected location. ^{99m}Tc pertechnetate showed features in keeping with sublingual thyroid with large areas of cystic photopaenia. No thyroid tissue demonstrated elsewhere. He was diagnosed with lingual thyroid lymphoma with hypothyroidism and prescribed with T Levothyroxine 100mcg OD.

Option for tracheostomy was given prior to commencement of chemotherapy, however patient refused. He developed acute upper airway obstruction during the course of chemotherapy and he succumbed.

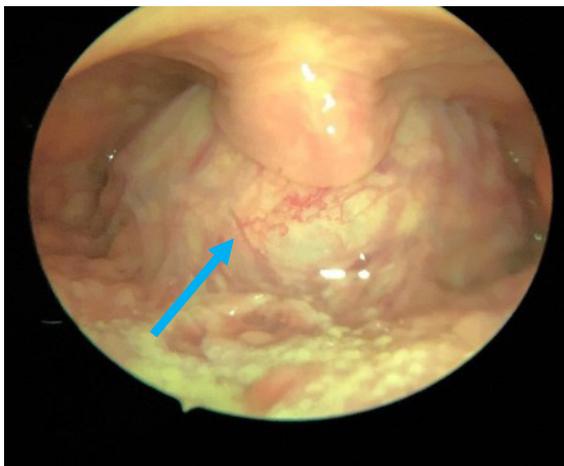


Figure 1: Arrow shows a large cystic mass occupying base of tongue and vallecula measuring 5x5 cm.



Figure 2: Arrow shows a large, well demarcated, hypodense lesion measuring 4.9x5.7cm.

Discussion

Ectopic thyroid first described and reported by Hickmann in 1869 [4]. The reported incidence was 1 in 100 000 cases which denotes the rarity of the condition [1,5]. Ectopic thyroid is defined as presence of thyroid gland other than its usual location. During embryological development, presence of thyroid gland

tissue is situated in the foramen caecum will descend slowly to its prelaryngeal position however due to unknown pathogenesis; thyroid gland will not be at the original site. The most common location for an ectopic thyroid is lingual (90%) whereas other locations could be in the submandibular region, thyroglossal duct or trachea [1]. Females are more prone for this abnormality with occurrence ratio of male to female is 1:7 [2]. Lingual thyroid is more obvious in female during puberty, pregnancy or menopause when plasma TSH level increases leading to hypertrophy of thyroid tissue [2].

Clinical presentation may vary from dysphagia, throat discomfort, hoarseness and occasionally upper airway obstruction [5]. Montgomery review done in 1935 showed that 70% of patients with lingual thyroid have hypothyroidism as in this case, it could be because of absent normal thyroid tissue [2]. Inflammation, hypertrophy, adenoma and malignancy of thyroid gland can be seen in ectopic thyroid. Malignant transformation of ectopic thyroid is extremely rare. Most tumours have been papillary carcinoma while others could be follicular, Hurtle cell carcinoma or lymphoma. Thyroid lymphoma accounts to less than 5% of all thyroid malignancies [7]. Similar to ectopic thyroid disease, thyroid lymphoma is mostly seen in female. Autoimmune (Hashimoto's) thyroiditis is the predisposing risk factor in development of thyroid lymphoma [8]. Usually, normal thyroid gland does not have lymphoid tissue. However, under pathological circumstances, most of them can transform into non Hodgkin lymphoma of B cell. Rapidly growing and painless neck swelling is the most frequent symptom. Patients can have pressure symptoms as well like dysphagia, dyspnea, hoarseness, cough and stridor.

Fine Needle Aspiration Cytology (FNAC) is inadequate to establish the diagnosis of lymphoma with no intact architecture. However, it is an excellent tool to eliminate other diagnosis for example carcinoma. When FNAC is not diagnostic but the suspicion is high, a core biopsy, incisional biopsy or even thyroidectomy is needed [9]. In this case, we opted for core biopsy when initial FNAC only yielded atypical lymphoid cells. The Ann Arbor staging is used to stage thyroid lymphoma by using Computed Tomography (CT) scan or Positron Emission Tomography (PET) CT [3]. PET scan is useful in monitoring disease recurrence.

Treatment of thyroid lymphoma mainly relies on both chemotherapy and radiotherapy [3]. Chemotherapy can control distant dissemination of the disease, while radiation therapy can achieve local control of the lymphoma. Chemotherapy regime for thyroid lymphoma includes cyclophosphamide, doxorubicin, vincristine and prednisone (CHOP). Chemotherapy is frequently combined with radiotherapy. Radiotherapy is commenced after 3 to 6 cycles of chemotherapy [9]. The radiation field includes neck with upper mediastinum and treatment dose is moderate (30-50Gy) [9]. Surgery has limited role and remains highly controversial. Surgery can be proposed in localized intrathyroidal lymphoma (stage 1E Ann Arbor Classification). But, if surgical resection is incomplete, radiotherapy should be considered [9].

Prognosis depends on the histological grade of the tumour and the stage of the disease. Mucosa-Associated Lymphoid Tissue (MALT) lymphoma tend to have better prognosis than diffuse large B cell lymphoma or mixed histological subtypes [9]. Early or advanced stage MALT lymphoma responds better to treatment than those with diffuse large B cell lymphoma.

Conclusion

In conclusion, ectopic thyroid lymphoma is a rare entity. Proper and thorough investigation can be done in order to determine the staging and prognosis of disease. Treatment approach should be given according to the symptomatology. Elimination of life threatening conditions such as airway obstruction should take precedence. The risk and benefit of each treatment modality should be discussed thoroughly with the patient to achieve optimum result.

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