Dual Ectopic Multinodular Goiter: A Case Report

Masoumeh Saeedi 1, Mohammad Hossein Khosravi 2,3

1 Department of Otorhinolaryngology-Head and Neck Surgery, Faculty of Medicine, Baqiyatallah University of Medical Sciences, Tehran, Iran
2 Students’ Research Committee (SRC), Baqiyatallah University of Medical Sciences, Tehran, Iran
3 International Otorhinolaryngology Research Association (IORA), Universal Scientific Education and Research Network (USERN), Tehran, Iran

Abstract

Background: As the first endocrine gland to form during embryogenesis, Thyroid develops in the 7th embryonic week through migration from foramen cecum to its adult position, anterolateral of 2nd and 4th tracheal cartilages [1]. Ectopic Thyroid can be found anywhere along the migration path with or without co-existence of a normally located Thyroid. Ectopic thyroid glands at two different locations are so rare that only about 24 cases of dual ectopia have been reported. We present the first case of dual ectopic submandibular multinodular goiter.

Case Report: We describe a 35-year-old housewife who consulted us complaining two huge and gradually-growing swellings in right and left upper neck regions. In physical examination, two great well-defined masses, one in right and one in left submandibular regions, were seen which had firm formidability with no mobility, tenderness or evidence of inflammation. Imaging and fine-needle aspiration confirmed the diagnosis of dual ectopic submandibular multinodular goiter in our patient as the first case of occurrence.

Conclusion: Finally, we recommend ectopic thyroid to be considered in the diagnosis of laterally located and submandibular neck masses especially in the absence of normally located thyroid. Additionally, we confirmed the idea that not all the laterally-located ectopic thyroids are malignant.[GMJ.2016;5(2):98-102]

Keywords: Ectopic Thyroid; Nodular Goiter; Multinodular Goiter

Introduction

As the first endocrine gland to form during embryogenesis, Thyroid develops in the 7th embryonic week through migration from foramen cecum to its adult position, anterolateral of 2nd and 4th tracheal cartilages [1]. Ectopic Thyroid can be found anywhere along the migration path with or without co-existence of a normally located Thyroid. In 90 percent of cases, it is located in the base of the tongue and in the remaining 10 percent, it is identified with other anatomic locations such as larynx, pharynx, esophagus, mediastinum and submandibular. Multinodular goiter (MNG) is defined as the enlargement of the thyroid gland with formation of nodules which has a total prevalence of 4.7-37.3% worldwide [2]. Ectopic thyroid can be involved by the same pathologies as the normally located one such as thyrotoxicosis or malignancy as well as MNG.
Ectopic thyroid glands at two different locations are so rare that only about 24 cases of dual ectopia have been reported [3]. We present the first case of dual ectopic submandibular MNG. Informed consent was obtained from patient included in the study.

**Case Presentation**

We describe a 35-year-old housewife who consulted us complaining two huge and gradually-growing swellings in right and left upper neck regions. During pregnancy and lactation period, the swellings had a significant progress. She was clinically euthyroid except with a history of constipation during pregnancy. In physical examination, two great well-defined masses; one in right and the other in left submandibular regions, were seen which had firm formidability with no mobility, tenderness or evidence of inflammation (Figure-1). Thyroid function test (TFT) was among normal ranges (TSH=2.27 mIU/L). Past medical history of the patient revealed no evident disease or hospitalization except one cesarean section surgery. Gastrointestinal cancer in her father was the only positive family history.

After physical examination, the patient was admitted for further evaluations. Axial neck computed topography (CT) scan, magnetic resonance imaging (MRI) (with and without contrast) and sonography were requested for her as well as fine needle aspiration (FNA). CT scan showed a huge lobulated mass with defined margin sized 130×60 millimeters with severe heterogeneous enhancement (Figure-2). Normal thyroid tissue was not seen in CT scan and the mentioned mass was suggested to be from thyroid origin based on CT scan report. Ultrasound evaluation revealed a double-lobe heterogeneous mass sized 130×60 millimeters with pressure effect on regional elements. Normal thyroid tissue was not seen in its anatomic place by ultrasound either. MRI revealed a very large and well circumscribed bilateral mass lesion sized 135×55 millimeters with heterogeneous signal on all sequences and mass effect on anterior side of pharynx at superior anterior part of neck in-

![Figure 1. Right and left submandibular swellings in the patient](image1)

![Figure 2. Axial image of contrast-enhanced CT scan showing the localization of left and right submandibular goiter (stars)](image2)
indicating intense enhancement on post Gado-
linium images. Normal thyroid gland tissue
was not seen in MRI. FNA was suggestive of
hyperplastic multinodular goiter. Radioactive
iodine uptake test (RAIU) showed an uptake
similar to two huge thyroid lobes.

Based on the aforementioned tests and imag-
ing, the patient was candidate for surgery with
diagnosis of MNG. Patient underwent midline
neck incision from left sternocleidomastoid
(SCM) muscle to right SCM. SCM muscle
was elevated from the mass which was stuck
to the hyoid bone by an isthmus. Therefore,
the body of hyoid bone was removed with
whole mass, which spread to the floor of the
mouth, by Sistrunk method. Extracted masses
(Figure-3) were sent for pathology assessment
which confirmed the ectopic multinodular
goiter.

Three hours after surgery, the patient had nau-
sea associated with one attack of vomiting.
The vital signs were stable. The day after sur-
gery, Calcium and Phosphorus levels were 9.5
and 3.1 mg/dl, respectively. Also, procalcite-
tonin (PCT) activity was reported 0.24 %. The
patient was discharged 3 days after surgery
with appropriate general condition. Patient
was referred to Endocrinologist for Thyroid
hormone therapy. The three-year postopera-
tive follow up was eventless.

Discussion

As the most common Thyroid development
disorder, ectopic Thyroid has an incidence
of 48-61%, the real incidence has remained
unknown though [4]. They are mostly asym-
ptomatic since cadaver studies have suggested
that 7-10% of adults may have thyroid tissue
along the embryonic migration pathway [5].
Moreover, thyroid tissue has been found in pa-
rotid gland, trachea, mediastinum, heart, lung,
duodenum, adrenal gland and even a more far
location, uterus [6]. Submandibular region is
another extremely rare reported location of
aberrant thyroid tissue [1, 7]. Ectopic thyroid
glands at two different locations are so rare
that only about 24 cases of dual ectopia have
been reported [3]. Our case presented with a
dual ectopic submandibular ectopic thyroid
tissue.

Ectopic thyroids are able to play the role of
a normal one; so they are potentially at the
risk of similar thyroid pathologies such as in-
flammation, thyrotoxicosis, malignancy, cal-
cification and MNG as in our case who was
diagnosed to have dual ectopic MNG. Multi-
nodular goiter is a slowly-progressive benign
disease which has different clinical manifesta-
tions during its course, a dominant nodule
at earlier and mass effect of enlarged goiter at
later stage [2].

Ectopic thyroid tissue in lateral neck was first
reported in the 18th century and further re-
ported cases were diagnosed to be malignant.
Thus, previous data recognized laterally-lo-
cated neck masses as “lateral aberrant thyroid
tumors” because they were considered to be either a metastasis from thyroid carcinoma or primary tumors of aberrant thyroid [8]. Albeit, a considerable number of laterally situated benign thyroid ectopia have been reported so far such as our case who was diagnosed with MNG. Ectopic thyroid may present as laterally-located mass with or without a normal eutopic thyroid as in our case [4]. Kousta et al. have suggested that Ectopic thyroid tissue should be considered in the diagnosis of a cervical mass even in the presence of a eutopic thyroid gland [6]. Thyroid ectopia may be associated with hypo-function or hyper-function of thyroid. However, thyroid function tests were within normal limits in our patient as in Akamnu et al. case [4].

Thyroid ectopia will clinically manifest and be diagnosed in periods of physiologic stress and demand of thyroid hormones as in menopause and adolescence [9]; as the masses had significantly grown during pregnancy in our patient.

Agrawal et al. reported a 13-year old male complaining of swelling in the right upper neck for 7 months and normal routine laboratory data. Physical examination revealed a 4×8 cm well-defined and non-tender submandibular mass. The patient underwent surgical excision of the mass and pathologic evaluation showed MNG[10]. Akanmu et al. reported a 34-year-old female with three left-sided neck masses which was diagnosed to be an enlarged multinodular left lobe of thyroid [4]. Although submandibular ectopic thyroid is rarely seen, it should be considered as a differential diagnosis of neck lumps. Furthermore, risk of metastasis or primary malignancy must be taken into account in such patients especially when there is a eutopic functional thyroid [7], as previous reports presented neck lumps were thought to be ectopic thyroid tissue but later found to be thyroid carcinoma metastases [1, 7].

To the best of our knowledge, the presented case is the first case of dual ectopic MNG. Regarding ectopic thyroid, location, size and the presence of symptoms or complications are determinative factors in treatment. The indications of treatment for MNG are suspected or confirmed malignancy, goiters with pressure effect, thyrotoxicosis, irritative symptoms or cosmetic concern [2]. Aside from mass effect reported by ultrasound, the chief complaint of our case was cosmetic concerns. Nearly all of the previous studies have suggested surgical excision of the mass in such patients as the treatment of choice [6, 10].

**Conclusion**

In conclusion, we described the first case of occurrence of dual ectopic submandibular multinodular goiter. We recommend ectopic thyroid to be considered in the diagnosis of laterally located and submandibular neck masses especially in the absence of normally located thyroid. Additionally, we confirmed the idea that not all laterally-located ectopic thyroids are malignant.

**Conflict of Interest**

Authors declare that they have no conflict of interest.