

Triple ectopic thyroid: A rarely described entity

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ABSTRACT

Introduction: Triple ectopic thyroid is a rarely described entity with only eight previous cases reported in the literature. This case is the first-described discovery of triple ectopic thyroid in a middle-aged male.

Case Report: A 46-year-old male with a past medical history of hypothyroidism and cannabis use disorder presented to his primary care physician with a globus sensation and left cervical lymphadenopathy for two weeks. A computed tomography (CT) scan of the neck with contrast identified three high attenuation areas: the first in the midline floor of the mouth extending posteriorly to the base of the tongue, the second at the base of the tongue, and the third in the midline of the neck just beneath the hyoid bone. Thyroid tissue was not identified in the orthotopic location. A follow-up thyroid uptake and scan confirmed that each of these high attenuation areas was also iodine avid, consistent with thyroid tissue.

Conclusion: The few cases of triple ectopic thyroid have mainly been discovered in female children with the most commonly described locations being the lingual (88%) and suprahyoid (88%) regions. Most patients were symptomatic with midline neck swelling. Overall,

we describe the first case of triple ectopic thyroid in a middle-aged male and the first patient with ectopic thyroid located in the midline mouth.

Keywords: Computed tomography of thyroid anomalies, Hypothyroidism, Lingual thyroid, Thyroid anomalies, Thyroid diseases, Thyroid dysgenesis, Thyroid gland ectopic thyroid, Thyroid uptake and scan of thyroid anomalies, Triple ectopic thyroid

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INTRODUCTION

During embryogenesis, the thyroid originates from the foramen cecum at the base of the tongue and finishes migrating toward its pre-tracheal position by the seventh week of gestation. This embryological process can arrest at any point, resulting in ectopic thyroid tissue. Generally, if ectopic thyroid tissue is present, there is only one ectopic site, rarely two. Exceedingly rare, with only eight cases reported in the literature, is triple ectopic thyroid [1–8]. The cases previously reported in the literature were mainly discovered in female children. In this case report and literature review, we present an extremely rare case of triple ectopic thyroid in a middle-aged male.

CASE REPORT

A 46-year-old male with a past medical history of hypothyroidism and cannabis use disorder presented to his primary care physician with a globus sensation and

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left cervical lymphadenopathy for two weeks. A computed tomography (CT) scan of the neck with contrast identified three high attenuation areas: the first in the midline floor of the mouth extending posteriorly to the base of the tongue (1.5×2.2 cm) (Figure 1), the second at the base of the tongue ($1.4 \times 1.8 \times 3.0$ cm) (Figure 2), and the third in the midline of the neck just beneath the hyoid bone (1.3 cm) (Figure 3). Thyroid tissue was not identified in the orthotopic location (Figure 4). A follow-up thyroid uptake and scan confirmed that each of these high attenuation areas was also iodine avid, consistent with thyroid tissue (Figure 5). Notably, there was no uptake in the normal orthotopic thyroid location.

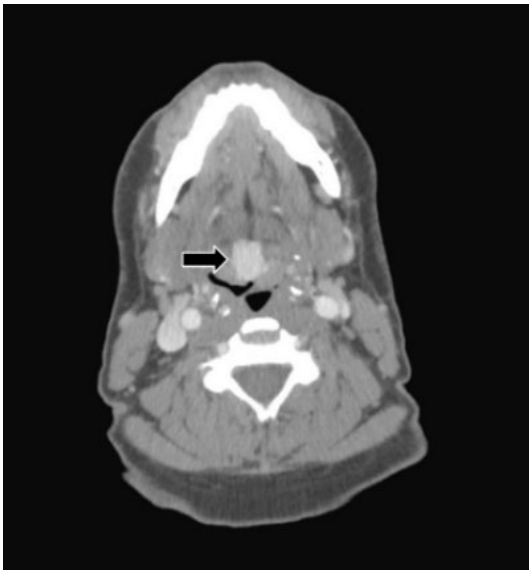


Figure 1: Ectopic thyroid tissue located on the midline floor of the mouth extending posteriorly to the base of the tongue (1.5×2.2 cm).

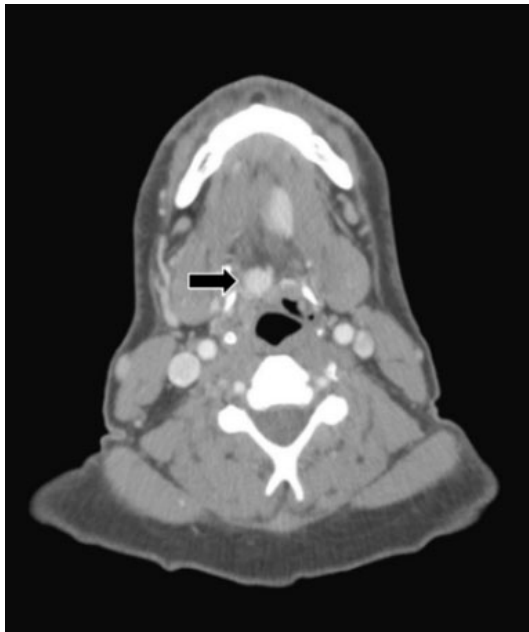


Figure 2: Ectopic thyroid tissue located at the base of the tongue ($1.4 \times 1.8 \times 3.0$ cm).



Figure 3: Ectopic thyroid tissue located in the midline of the neck just beneath the hyoid bone (1.3 cm).

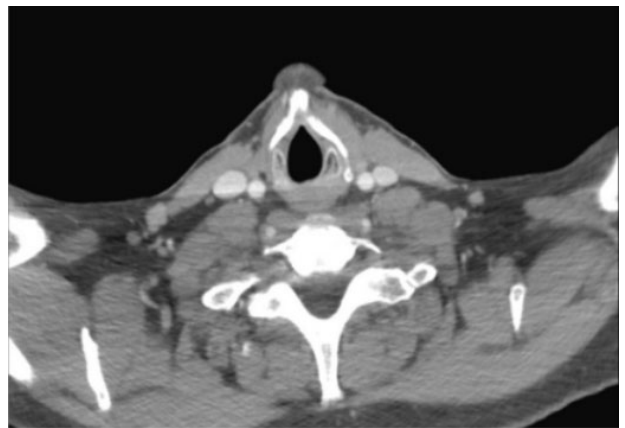


Figure 4: Lack of thyroid tissue at the normal orthotopic location.



Figure 5: Thyroid uptake and scan confirming the three iodine-avid areas consistent with ectopic thyroid tissue. Notably, there is no uptake in the normal orthotopic thyroid position.

In regard to the patient’s hypothyroidism, he was diagnosed 12 years prior to presentation on routine screening and has been prescribed an increasing dose of levothyroxine replacement therapy since then. He did not have any thyroid imaging done at the time of diagnosis.

In regards to the patient’s symptoms, his left cervical lymphadenopathy and globus sensation were evaluated and not thought to be related to the ectopic thyroid tissue.

DISCUSSION

Thyroid tissue originates from the foramen cecum at the base of the tongue during embryogenesis. As the endodermal cells proliferate, the thyroid diverticulum is created which begins migrating toward its pre-tracheal position [9]. The thyroid reaches its normal anatomic location by the seventh week of gestation [9]. The thyroglossal duct remains along the thyroid’s tract of descent until week ten of gestation when it degenerates [9]. This embryological process can arrest at any point resulting in ectopic thyroid tissue. The prevalence of

ectopic thyroid tissue is approximately 1 per 100,000–300,000 people, though the prevalence increases dramatically to nearly 7–10% of the population when looking at autopsy studies [10]. The most common location for ectopic thyroid tissue is the foramen cecum (lingual ectopic thyroid) which comprises 90% of ectopic thyroid cases [10]. Other less common locations have been reported, mainly along the tract of the thyroglossal duct, including submandibular, suprahyoid, and infrahyoid [10]. Ectopic thyroid tissue has also been reported in areas inferior to the normal orthotopic thyroid location including intrathoracic (mediastinum, lung, and heart), struma ovarii, adrenal gland, duodenum, pancreas, and intestines [10]. In cases of dual ectopic thyroid tissue, the areas involved most commonly included the lingual and subhyoid sites [10].

Even rarer than dual ectopic thyroid is triple ectopic thyroid. In fact, there are only eight previously reported cases in the literature [1–8]. Here, we will review the previously reported cases including the most common location of the ectopic thyroid tissue as well as the age, sex, symptomatology, and thyroid function of each of these patients. This is also summarized in Table 1.

Table 1: Summary of previously reported triple ectopic thyroid cases

Study	Age (years)	Gender	Ectopic thyroid location	Symptoms	Thyroid status	Orthotopic thyroid present?	References
Triple ectopic thyroid detected on SPECT/CT	11	Female	Lingual, suprahyoid, infrahyoid,	Midline neck swelling, delay in growth milestones	Euthyroid: TSH 4.12 uIU/mL, FT4 1.24 ng/L, FT3 3.9 pg/mL	No	[1]
A rare case of triple thyroid ectopia	42	Female	Lingual, suprahyoid, infrahyoid	Asymptomatic, found incidentally during unrelated imaging study	Biochemical profile unavailable	No	[2]
Rare developmental abnormalities of thyroid gland, especially multiple ectopia: A review and our experience	20	Female	Lingual, suprahyoid, and infrahyoid	Midline neck swelling	Subclinical hypothyroidism (biochemical profile unavailable)	No	[3]
Triple ectopic thyroid on pertechnetate scintigraphy	5	Female	Lingual, suprahyoid, pretracheal	Midline neck swelling	Subclinical hypothyroidism: TSH 7.55 uIU/mL, FT4 6.62 ug/dL, FT3 1.34 ng/mL	No	[4]
Triple ectopic thyroid	16	Female	Lingual, suprahyoid, infrahyoid	Midline neck swelling	Euthyroid (biochemical profile unavailable)	No	[5]
Triple ectopic thyroid: A case report and review of literature	10	Female	Lingual, suprahyoid, pretracheal	Midline neck swelling	Euthyroid at presentation (biochemical profile unavailable). Five months later, the patient developed hypothyroidism: TSH 5.71 uIU/L, FT4 0.7 ng/dL	No	[6]

Table 1: (Continued)

Study	Age (years)	Gender	Ectopic thyroid location	Symptoms	Thyroid status	Orthotopic thyroid present?	References
Multiple ectopic thyroid masses in a hypothyroid child	6	Male	Lingual, suprahyoid, cricoid cartilage	Midline neck swelling	Hypothyroidism: TSH 53 IU/ mL	No	[7]
Lateral cervical ectopic thyroid masses with eutopic multinodular goiter: An unusual presentation	34	Female	Laterally	Midline and lateral neck swelling	Euthyroid: TSH1.1 mU/ mL, FT4 50 ng/mL, FT3 1.3 ng/mL	Yes	[8]

In the reported cases of triple ectopic thyroid, the most common locations were the lingual (88%) and suprahyoid (88%) regions [1–7]. Less common locations included the infrahyoid (50%), pre-tracheal (25%), and cricoid cartilage (13%) areas [1–7]. Lastly, one patient only had ectopic thyroid lateral to the orthotopic thyroid [8]. In addition to lingual (Figure 1) and infrahyoid (Figure 2) areas, our patient had ectopic thyroid in the midline mouth (Figure 3) which has not previously been described and would seem quite unusual given the embryological pathway in normal thyroid development.

In terms of age, the known cases of triple ectopic thyroid ranged from 5 years old to 42 years old [1–8]. The average age was 18 years old. In terms of sex, seven of the eight known cases occurred in females [1–6, 8]. The one previous male patient with triple ectopic thyroid was 6 years old at the time of diagnosis [7]. Therefore, our patient is not only the oldest described patient with triple ectopic thyroid at age 46, but he is also only the second described male patient and first described adult male patient.

In terms of symptomatology, 87.5% (seven out of eight) patients experienced neck swelling, mainly midline [1, 3–8]. The last remaining patient was identified incidentally due to another imaging study [2]. Our patient experienced midline neck swelling which is likely related to the ectopic thyroid tissue. The globus sensation is thought to be unrelated to the ectopic thyroid tissue.

Finally, there was variation in the thyroid function of each of these patients at diagnosis. There was no information on the thyroid function status of one patient [2]. Of the remaining seven patients, three were euthyroid, two had subclinical hypothyroidism, one was hypothyroid, and the remaining patient was euthyroid upon diagnosis and subsequently developed hypothyroidism within five months of diagnosis [1, 3–8]. Of note, only one patient had orthotopic thyroid tissue and she was euthyroid [8]. Our patient was hypothyroid and had been treated with increasing doses of levothyroxine for 11 years prior to diagnosis with triple ectopic thyroid. Given the early age of diagnosis of most of these patients, it is unclear at this time if hypothyroidism is perhaps a

common manifestation of triple ectopic thyroid as these patients age.

CONCLUSION

All in all, triple ectopic thyroid remains an exceedingly rare diagnosis. Our patient is currently the only adult male patient described in the literature and the first patient with ectopic thyroid located in the midline mouth.

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Author Contributions

Stephanie AC Pitts – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Pratima Sood – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Ronnie D Derrwaldt – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all

aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Guarantor of Submission

The corresponding author is the guarantor of submission.

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Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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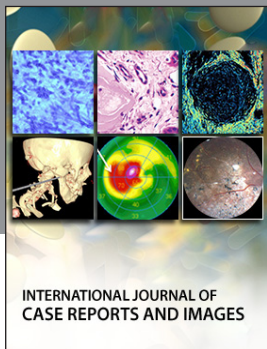
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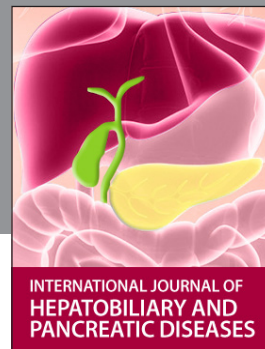
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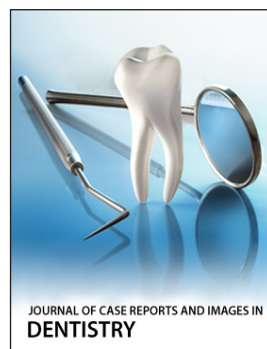
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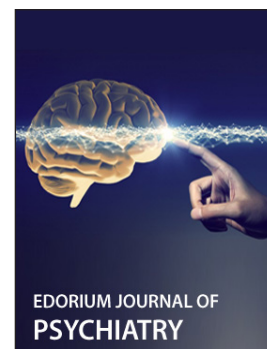
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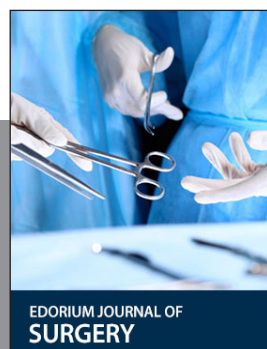
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