

Detection of Thyroid Isthmus Agenesis during Diagnostic Lobectomy: A Case Report

Ann. Ital. Chir., 2024 95, 4: 477–480
<https://doi.org/10.62713/aic.3306>

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The thyroid gland is an endocrine organ comprising two lobes connected by an isthmus. Thyroid isthmus agenesis (TIA) is a rare anatomical anomaly, which has only been documented in a limited number of case reports. This report presents the case of a 49-year-old female patient who was diagnosed with intraoperative TIA during diagnostic thyroid lobectomy. The patient was arranged for a diagnostic lobectomy of the right thyroid lobe after identifying an atypical nodule of undetermined significance, in the right lobe, which had been confirmed with two distinct biopsies. The patient was discovered to have isthmus agenesis during a neck exploration performed under general anesthesia. Owing to its rarity, the precise clinical and anatomical characteristics of TIA have not been defined, and its underlying cause is still not completely understood. The detection of TIA warrants the considerations of any other accompanying diseases. Prior identification and assessment of surgical approach are crucial for ensuring a secure surgical procedure and mitigating the risk of surgical complications. Nevertheless, caution must be exercised if TIA is detected during operation because preoperative identification is not always feasible. In summation, additional patient reports and studies are required to uncover the underlying cause of this pathological condition.

Keywords: thyroid; thyroidectomy; lobectomy; isthmus; agenesis; atypia of undetermined significance/follicular lesion of undetermined significance

Introduction

The thyroid gland is an endocrine organ comprising two lobes connected by an isthmus. Thyroid gland starts to develop with the invagination of endodermal cells in the ventral floor of the primitive pharynx in the fourth intrauterine week. This organ is vulnerable to a diverse range of morphological variations and developmental anomalies, including hypoplasia, ectopy, hemiagenesis, and agenesis. Thyroid isthmus agenesis (TIA) is a rare anatomical anomaly, which has been sparingly documented in the literature [1, 2]. Given its rarity, the exact cause of TIA is not completely understood. However, there have been reports indicating a possible link between TIA and genetic factors, particularly those involving thyroid transcription factor-1 (*TITF1*) and thyroid transcription factor-2 (*TITF2*) genes and mutations in chromosome 22. The exact incidence of this condition is currently unknown due to the limited number of case reports documented in the literature. However, based on the cadaver studies, the incidence rate of TIA is estimated to range between 0.5% and 10% [2, 3].

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This report presents the case of a patient who were found to have TIA during diagnostic lobectomy.

Case Presentation

This study was conducted in accordance with the Declaration of Helsinki. Signed informed consent was obtained from the patient after she agreed to publish her clinical case. The conduct of this case study has been approved by the ethics committee of Sultan II. Abdulhamid Han Training and Research Hospital, which demanded the acquisition of informed consent of the patient.

A 49-year-old female patient with a medical history of multinodular goiter was found to have isthmus agenesis detected during diagnostic lobectomy of the right thyroid lobe. No palpable thyroid nodule was detected in the patient during physical examination. Neck ultrasonography (USG) revealed multiple nodules, the largest of which was in the right lobe of the thyroid gland; no nodule was detected in the left lobe. Fig. 1 shows the patient's preoperative neck ultrasonography (USG) image.

Several thyroid function parameters of the patient were tested. The levels of both thyroid-stimulating hormone (TSH) = 3.02 mIU/L (normal value: 0.54–4.31 mIU/L) and FT₄ = 1.07 ng/dL (normal value: 0.85–1.70 ng/dL)

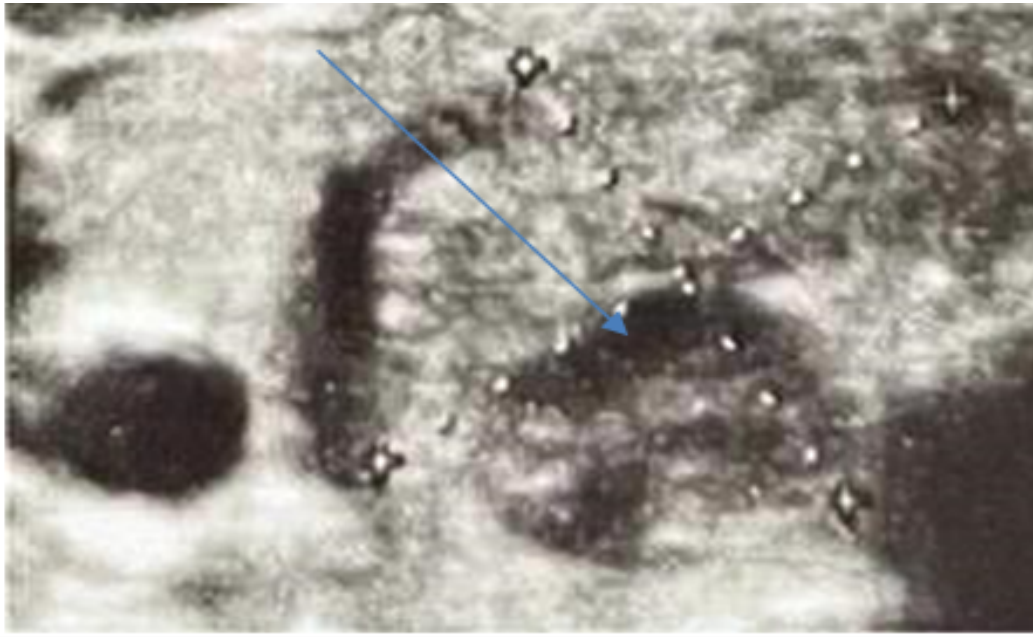


Fig. 1. Preoperative neck ultrasonography image. The blue arrow indicates the nodule in the right lobe of the thyroid gland.



Fig. 2. Intraoperative neck exploration conducted on the patient.

were within the normal range [4]. The levels of FT4 and TSH were measured with Roche Cobas analyzers (V. 6000, Roche Diagnostics, Mannheim, Germany).

The patient was arranged for a diagnostic lobectomy of the right thyroid lobe after detecting an atypical nodule of undetermined significance, in the right lobe, which had been

validated with two distinct biopsies. Isthmus agenesis was discovered during a neck exploration performed on the patient under general anesthesia. The patient's intraoperative neck exploration image is shown in Fig. 2.

Right lobectomy was carried out under neuromuscular nerve monitoring guidance. The patient was discharged without any complications on the first postoperative day. In addition, histopathological examination of the patient's right lobectomy specimen revealed no evidence of malignancy but the presence of benign nodular hyperplasia.

Discussion

Given the rarity of TIA, the clinical community has been facing a dearth of case reports about this morphological anomaly, which provides insufficient knowledge support to clinicians, rendering preoperative detection of TIA mostly unfeasible. Adding to the challenges of preoperative detection of TIA is often the its asymptomatic nature, which explains why this pathological condition is, in most cases, detected incidentally because of the comorbid thyroid pathologies [1]. Preoperative detection and surgical strategy determination are crucial to ensure a safe surgical procedure and avoid any potential complications. In the event of elevated TSH levels, thyroid scintigraphy can be employed to facilitate diagnosis. In addition, TIA can be diagnosed by radiological means such as USG, computed tomography (CT) and magnetic resonance imaging (MRI), or without involving invasive surgical methods. Upon the detection of TIA, differential diagnosis should be performed to rule out the possibility of autonomous thyroid nodule, thyroiditis, primary carcinoma, neoplastic metastasis, and infiltrative diseases such as amyloidosis. It is important to note that TIA can be associated with ectopic thyroid tissue or parathyroid hyperplasia [2, 5].

The existing literature suggests that the arteria thyroidea ima (ATI) may contribute to the occurrence of TIA, but further research is required to fully understand the underlying causes of TIA. This is because the usage of CT scans is not sufficient for an in-depth evaluation of ATI [6]. This possibility suggests that greater caution should be exercised to mitigate the risk of perioperative bleeding. Despite of the incomplete characterization of TIA etiologies, a previous study has illustrated the genetic basis in developmental anomalies, specifically the involvement of thyroid transcription factor 1 (*TITF1*) and thyroid transcription factor 2 (*TITF2*) gene as well as chromosome 22 anomalies [2]. The management of TIA also presents huge challenges because there is currently no clear definition of the specific clinical and anatomical characteristics of TIA, given the scarcity of the existing literature [6]. Hence, more case reports, clinical trials, and radiological studies in this regard are required.

Conclusion

Owing to the undefined clinical and anatomical characteristics, a rigorous differential diagnosis should be performed

if the TIA is speculated. Caution must be exercised if TIA is detected intraoperatively because preoperative identification is not always feasible. To enhance surgical safety and mitigate the risk of surgical complications, it is crucial to perform preoperative identification and evaluation of surgical approach to be utilized.

Availability of Data and Materials

The data that support the findings of this study are available from the corresponding author, upon reasonable request.

Author Contributions

UK: Study design, analysis, interpretation, writing the article, critical revision of the article, and literature review. MGD: Literature review, English editing, critical revision. YKK: Literature review, critical revision. AFM: Data collections, literature review. MS: Literature review, critical revision. All authors: Analysis and interpretation of data. All authors revised the manuscript critically for important intellectual content. All authors read and approved the final manuscript. All authors have participated sufficiently in the work and agreed to be accountable for all aspects of the work.

Ethics Approval and Consent to Participate

Signed informed consent was obtained from the patient included in this case report and is available upon request. The Declaration of Ethics, which requires only the patient's informed consent because it is retrospective and complies with CARE rules, has been exempted by Sultan II. Abdulhamid Han Training and Research Hospital. The study was conducted in accordance with the Declaration of Helsinki.

Acknowledgment

Not applicable.

Funding

This research received no external funding.

Conflict of Interest

The authors declare no conflict of interest.

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