



Case report

Ectopic thyroid carcinoma in the mandible with normally located goiterous thyroid gland: A case report



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ABSTRACT

Background: The presence of ectopic thyroid tissue is a developmental disorder that affects about one per 100,000–300,000 people. It is extraordinarily rare for ectopic thyroid to be out of the lines of its descent. This case is the first to report a cancerous ectopic thyroid in the mandibular bone with a goitrous orthotopic thyroid gland.

Case presentation: 80-year-old female of free medical background presented to our clinic with painless left mandibular swelling that recent increase in size after tooth extraction. Surprisingly the diagnosis of ectopic thyroid tissue with cancerous follicular cells was reached upon histological analysis. The goitrous thyroid gland was found in its expected cervical location, and the patient was euthyroid.

Conclusion: Because this diagnosis extremely rare, the possibility of ectopic thyroid carcinoma in cases of a pathological mass in the mandible must always be carefully thought out. A high degree of suspicion is essential as the unusual presentation could take.

1. Introduction

Ectopic thyroid tissue is the presence of thyroid tissue in locations other than its anterior neck region anterolateral to 2-5th tracheal rings. The exact cause is not fully known yet, but an association with defects in developmental morphogenesis and differentiation was reported [1]. Its prevalence is about 1 per 100,000–300,000 people, and 70–90 % of them lack other thyroid tissue. The most commonly reported location accounting for 90 % of cases, is at the base of the tongue [2]. Other described sites include lingual and sublingual, tracheal, submandibular, lateral cervical region, carotid, axillary, iris, pituitary, cardiac, aortic and pulmonary, esophageal, duodenal, gallbladder, gastric, pancreatic, mesenteric, porta hepatis, adrenal, ovaries, tubaric, uterine and vaginal [1]. Very uncommon reports of dual, triple and multiple ectopias with or without normal orthotopic thyroid [3].

The orthotopic thyroid gland usually coexists; in most cases, the patients are euthyroid. All pathological events that could affect the thyroid gland can be present in the ectopic.

Primary thyroid carcinoma in ectopic tissue is exceptionally

uncommon and challenging to be differentiated from metastatic lesions.

Scintigraphy, using Tc-99m, I-131, or I-123, is the most important diagnostic tool, although U.S., CT, MRI and thyroid function tests should be considered in designating the extension and location of ectopic tissue. Fine needle aspiration cytology (FNAC) dramatically assists in confirming the diagnosis of ectopic thyroid. It is a modality of great value to differentiate between a benign and a malignant lesion [4].

Most authors agree that the best treatment strategy for ectopic thyroid is linked to the patient's age, localization, local symptoms, malignancy, surgical and anesthesiological risk management, and thyroid functional status [2].

.Our case reports a malignant ectopic thyroid tissue in the mandibular bone with an abnormal thyroid gland in its standard anatomical location [5]. As a malignant ectopic thyroid tissue in the mandible, this is the first case to be reported in the literature.

This work has been reported in line with the SCARE criteria [6].

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2. Case presentation

80-year-old female of free medical background presented to surgery outpatient clinic of Military Hospital Omdurman, Sudan with painless left mandibular swelling that gradually increased in size over 18 months after a left lower first molar tooth extraction procedure with no other complaints. Not on any medication and no reported drug allergy and with no family history of similar conditions or malignancies. She is from a low socioeconomic background. On physical examination, there was a left side mandible swelling, 8 × 6 cm, oval in shape, hard, smooth surface, tender, immobile and with normal skin over (Fig. 1). Intraorally, multiple teeth were lost on the left side with a swelling extended into both buccal and lingual sides (Fig. 2).

The patient was then referred to the Oral and Maxillofacial Surgery Department for assessment. After history and examination, the orthopantomogram showed destruction of the left mandibular bone body and multiple lower teeth losses (Fig. 3). Initial diagnosis of ameloblastoma, odontogenic keratocyst and the malignant lesion was suspected. Incisional biopsy was taken through an intraoral approach, and the result of the histopathological assessment was Consistent with ectopic thyroid tissue.

After that, further workup was started, including a thyroid function test that revealed a euthyroid state. The left mandibular lesion ultrasonography showed a heterogenous left jaw swelling 5.3 × 4 with multiple nodular appearance, cystic changes, central vascularity and smooth outlines. Neck ultrasonography revealed a thyroid gland with an enlarged right lobe, a cystic nodule measuring 2.3 × 2 cm, and a normal left lobe. There was no evidence of any significant cervical lymphadenopathy.

A computed tomography scan revealed an expansile lytic lesion in the body of the left mandible, associated with a large soft tissue mass. The soft tissue mass showed inhomogeneous enhancement after I.V. contrast injection, and it was concluded that these findings suggest osteosarcoma of the left mandible (Fig. 4). It also showed calcification at the root of the right thyroid gland lobe.

Accordingly, thyroid gland FNAC was taken, and the smears showed numerous clusters of follicular cells, mixed inflammatory cells and red blood cells. They were interpreted to be suggestive of follicular neoplasm and hyperplastic goitre. A thyroid scan revealed a mildly enlarged thyroid gland normally located with a cold nodule occupying the lateral part of the right lobe (Fig. 5). However, it did not elaborate on the existence of another ectopic Uptake.

All blood tests, chest radiograph, echocardiography and brain C.T. appeared normal.

The patient underwent a mandibular mass excision and total thyroidectomy surgeries in the same session in the military hospital surgery department by experienced operators. The abnormal bone and underlying mass were excised through an intra-oral incision extended from the midline to the mandibular angle. It was a 5 × 4 cm encapsulated,



Fig. 1. Preoperative clinical finding of the mandibular swelling.



Fig. 2. Intra-oral clinical finding of the mandibular swelling extending into buccal and lingual sides.

brownish soft tissue mass which exhibited a firm consistency (Fig. 6). The posterior cortex of the mandibular bone was preserved, and there was no need for reconstruction. The histopathological assessment revealed a diagnosis of follicular ectopic thyroid carcinoma (Fig. 7). Total thyroidectomy was done due to the lack of a frozen section in our theatre. The isolated thyroid gland was 5 × 3 × 2 cm in dimensions exhibiting a right lobe nodular hard brownish area (Figs. 8; 9). Histologically diagnosis was reported as nodular colloid goitre (Fig. 10).

Postoperatively the patient recovered well, and it was an uneventful course until day four that we sadly confirmed the unfortunate patient's sudden death. The cause of death was unknown, and common post-operative cardiac complications are highly predicted. Although a moderate risk for pulmonary embolism was calculated, there was no timely access to further workup.

3. Discussion

When dealing with ectopic thyroid tissue, we must have extensive knowledge of its embryological origin. The abnormal tissue usually can be found in the path the thyroid takes to its final resting place. The thyroid develops from the floor of the pharynx, then descends to the pharyngeal gut and ultimately reaches the base of the neck [1]. Throughout this process, the gland remains connected to the tongue with a duct called the thyroglossal duct that degenerates and allows the thyroid to detach and reach its final normal midline plane. The tissue can also be found in the adrenal gland, liver and other rare cases [5].

Its prevalence is about one per 100,000–300,000 people, rising to 1 per 4000–8000 patients with thyroid disease. It may develop at any age, from 5 months to 40 years, but is most common at younger ages [7].

Generally, the ectopic thyroid tissue can be affected by the same diseases as the normally situated thyroid [8]. Primary thyroid carcinoma in ectopic tissue is exceptionally uncommon, however, and has been reported in cases of lingual thyroid, thyroglossal duct cyst, lateral aberrant thyroid tissue, mediastinal, and struma ovarii [9].

The common differential diagnoses for intra-oral mandibular swelling are ameloblastoma, odontogenic keratocyst and malignant masses. Moreover, various differential diagnoses exist through congenital, benign and malignant conditions.

Our case's unanticipated diagnosis of ectopic thyroid tissue was luckily reached with the incisional tissue biopsy. At that point, we already excluded that this could be the result of metastases, as a colloid goitre was found from the FNA previously done on the normally situated thyroid. We had to wait for the post-surgical pathology reports to reach this result, as determining whether the ectopic thyroid tissue was metastatic or a primary malignant lesion proved difficult.

In general, the most important diagnostic modality for ectopic thyroid tissue is thyroid scanning with technetium-99 m, a sensitive and specific imaging modality in diagnosing functioning ectopic thyroid

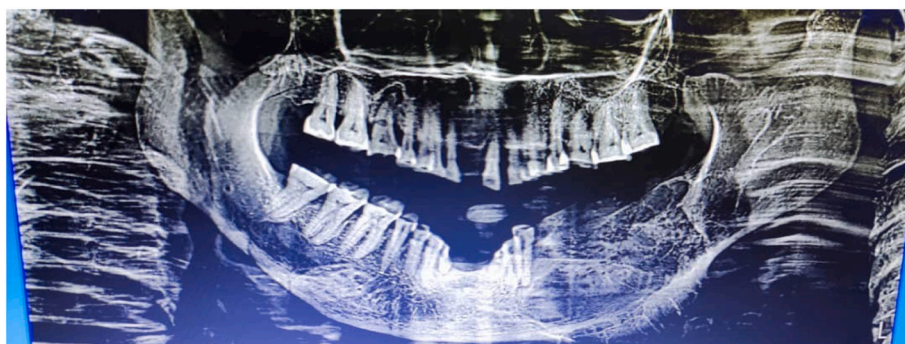


Fig. 3. The preoperative orthopantomogram shows large-scale destruction in the left mandible.

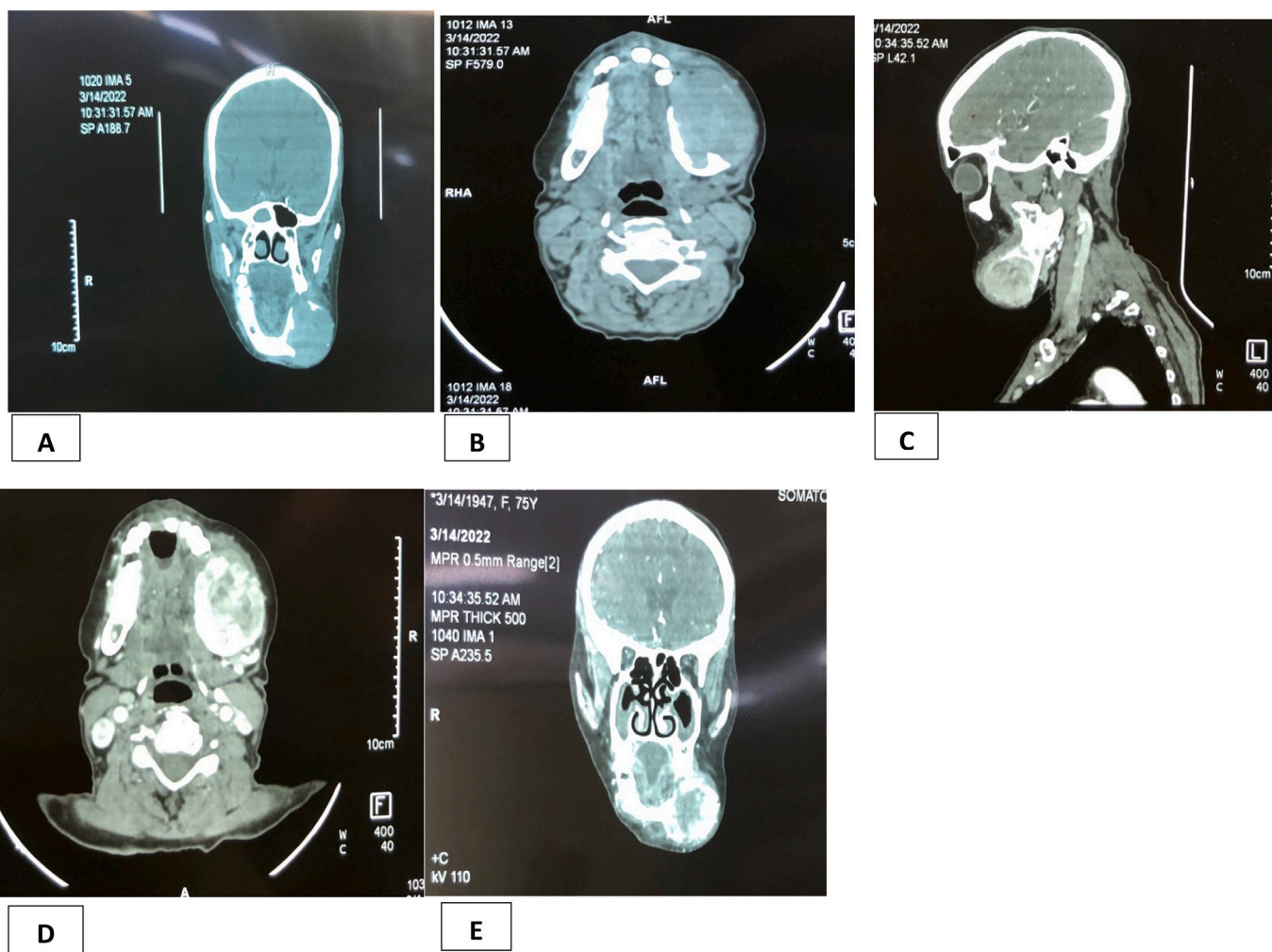


Fig. 4. The preoperative computed tomography scan.
 A. The coronal section shows a left mandible lytic lesion with soft tissue mass.
 B. Axial view.
 C. The sagittal section shows the left mandible soft tissue mass.
 D. Post-contrast axial view showing inhomogeneous enhancing lesion on the left side.
 E. Post-contrast coronal section.

tissues. Thyroid scanning importantly helps in confirming the presence of a normally located thyroid gland before any surgical intervention in ectopic tissue, so; it was the main elaborator of our patient orthotopic thyroid gland [10]. However, the normally located thyroid gland is absent in about 70 % of cases.

FNAC, USG, CT, and MRI have a significant role in defining the

extension and location of the ectopic thyroid gland.

The presence or absence of orthotopic functioning thyroid tissue is the core of diagnostic and therapeutic interventions. Although unfortunately, in our condition, suspicion of malignancy indicates the total excision of the gland and a plan of thyroxin replacement therapy [11].

Because of its different circumstances and rarity, there is no specific

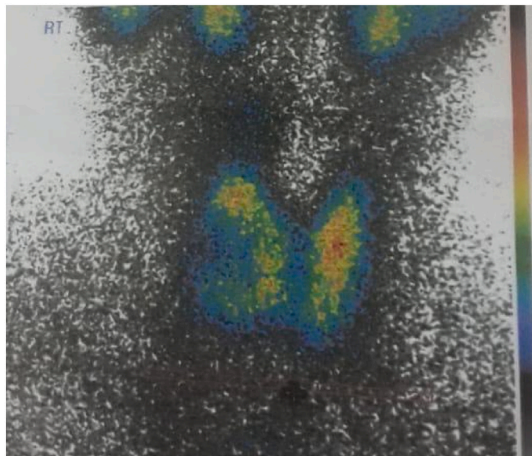


Fig. 5. A thyroid scan with Tc-99m reveals a mildly enlarged, normally located thyroid gland with a cold nodule in the right lobe.

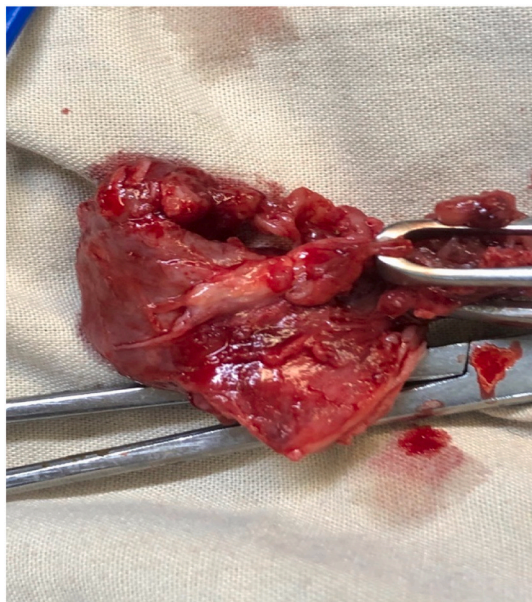


Fig. 6. Postoperative specimen showing the excised ectopic thyroid mass.

optimal treatment plan for ectopic thyroid tissue.

Although carcinomas in ectopias are rare, malignancy should always be suspected. As indicated in the literature, most malignant ectopic thyroid tissue is accompanied by a corresponding thyroid gland malignancy, and most are diagnosed incidentally after surgical excision.

Ectopic thyroid tissue must be differentiated from metastatic thyroid cancer intensively with malignant ectopia. As the ectopic thyroid tissue shows malignant follicular changes in our case, FNAC and tissue histopathological assessment of the orthotopic gland helped exclude metastatic cancer. Other distant Mets should also be excluded.

Patient risks and significant postoperative complications should be considered when considering surgical interventions.

4. Conclusion

Because it does not ordinarily to come to mind, it is a challenging diagnosis to reach. Thus, the surgeons must always consider the possibility of ectopic thyroid carcinoma in cases of a pathological mass in the mandible. A high degree of suspicion on imaging is essential as the unusual presentation it could take.

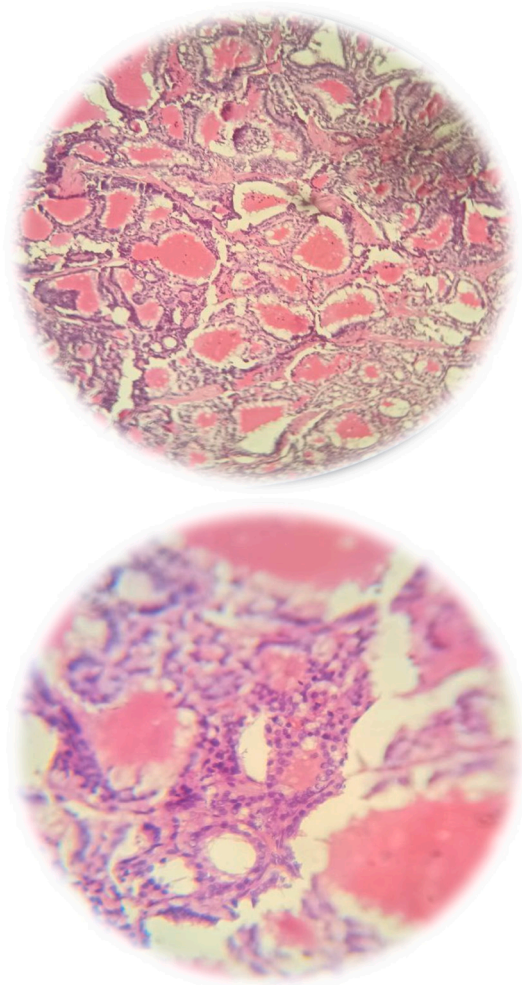


Fig. 7. Photomicrograph of the histologic section from the isolated ectopic thyroid tissue indicates follicular carcinoma. The lesions consist of different-sized follicles with a dense appearing colloid. Tumour cells are cuboidal with dark staining round nuclei with inconspicuous nucleoli. They are shown with low and high-power magnification using Hematoxylin and Eosin stain.

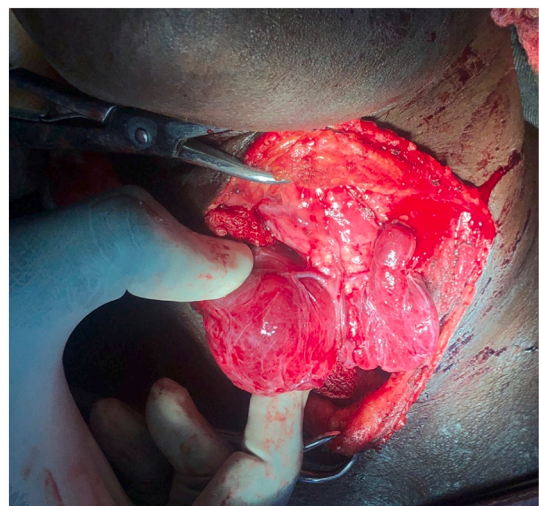


Fig. 8. Intraoperative view of the goitrous thyroid gland with right lobe large nodule.



Fig. 9. Postoperative specimen of the isolated thyroid gland with right lobe large nodule.

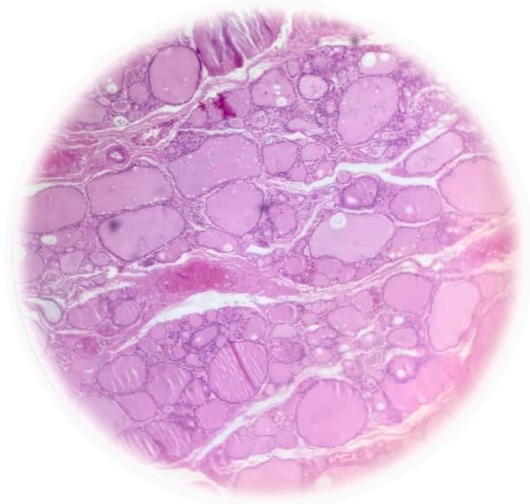


Fig. 10. Photomicrographs of a histologic section of the thyroid gland indicate colloid goitre using Hematoxylin and Eosin stain.

A benign thyroid gland should not exclude the existence of malignant ectopic thyroid tissue elsewhere.

Declaration of competing interest

The authors declare no conflict of interest regarding the publication of this paper.

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Ethical approval

Ethical approval was obtained from the Military hospital ethical committee.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Ali Mohamed: Study concept and Design and Supervision.

Mohamed Noon: Data curation, Writing-Original Draft Editing and case preparation.

Ahmed Ahmed: Case preparation. Writing_Original draft, and Editing.

Adam Najdelddin: Case preparation and Editing.

Basher Mohamed: Writing_Reviewing and Editing.

Research registration

N/a.

Guarantor

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