

'Forgotten' goitre after total thyroidectomy

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Background. 'Forgotten' goitre (FG) is a mediastinal portion of the thyroid found after total thyroidectomy. It is extremely rare.

Objectives. To report on 5 cases and review the literature.

Methods. We retrospectively reviewed all patients with retrosternal goitre at Mohammed V Military Teaching Hospital, Rabat, Morocco, from 1 January 2010 to 31 December 2020 and identified 5 patients who underwent surgery for forgotten mediastinal goitre during this period. Data on patient characteristics, time of the first thyroidectomy, imaging (chest radiograph, chest computed tomography (CT) scan and magnetic resonance imaging), surgical approach, pathological findings and postoperative complications were collected.

Results. Five patients (3 female and 2 male) with a mean age of 46.2 years (range 32 - 56 years) with FG had surgery. Two patients were asymptomatic and 3 patients had dyspnoea, 2 of whom also had dysphonia and 1 signs of hyperparathyroidism. No patient had a history of thyroid cancer. The average time between the first thyroidectomy and re-operation was 4.3 years. The diameter of the masses on CT scan was 7 - 12 cm. All the patients underwent sternotomy. Postoperative transitory left recurrent laryngeal nerve palsy occurred in 1 case. There were no postoperative deaths. Pathological examination of the mediastinal goitre confirmed multi-heteronodular thyroid hyperplasia in all cases.

Conclusion. FG is an extremely rare condition, which can be prevented with thorough preoperative imaging.

Keywords. Forgotten goitre, mediastinum, chest computed tomography, recurrent nerve palsy, sternotomy.

Afr J Thoracic Crit Care Med 2025;31(2):e762. <https://doi.org/10.7196/AJTCCM.2025.v31i2.762>

Study synopsis

What the study adds. 'Forgotten' goitre is a mediastinal portion of the thyroid found after total thyroidectomy. It is extremely rare. We report on 5 cases.

Implications of the findings. Thorough preoperative imaging of retrosternal goitre can prevent the occurrence of forgotten goitre and its associated morbidities. Sternotomy is usually required for reoperation, highlighting the importance of complete initial thyroidectomy.

'Forgotten' goitre (FG) is a mediastinal thyroid mass found after total thyroidectomy for retrosternal goitre. The frequency reported in the literature varies between 2% and 16% of cases of retrosternal goitre.^[1-5] A search using the terms 'forgotten goiter after thyroidectomy' and 'missed goiter after thyroidectomy' in the PubMed and Google Scholar databases showed that ~40 cases have been reported. Neck and chest computed tomography (CT) and/or chest magnetic resonance imaging (MRI) play an important role in the preoperative work-up of patients with FG to prevent complications. Preoperative imaging is also important before initial thyroidectomy to avoid missing thyroid tissue.

Methods

Between 1 January 2010 and 31 December 2020, 4 266 patients underwent thyroid surgery at Mohammed V Military Teaching Hospital, Rabat, Morocco. Of these, 167 (3.9%) had retrosternal goitre and were operated on by the thoracic surgery department. In the majority of these cases ($n=161$; 96.4%) a cervical approach was possible, while 6 patients (3.6%) required a sternotomy. Five cases of FG were identified. All these patients were referred from other

departments or hospitals. Data on age, gender, symptoms, initial pathological findings, hormone tests (free tri-iodothyronine, free thyroxine, thyroid-stimulating hormone (TSH)), radiological findings (chest radiograph, CT, MRI), surgical approaches, postoperative complications and hospital stay were collected from medical records. Laryngoscopy was routinely performed to assess recurrent laryngeal nerve function.

Results

The patients ranged in age from 32 to 56 years (mean 46.2), and 3 of them were female. The time between the thyroidectomy and discovery of the mediastinal mass ranged from 6 months to 8 years, with an average of 4.3 years (Table 1).

Symptoms were present in 3 patients, all of whom had dyspnoea secondary to tracheal compression, with dysphonia in 2 cases and signs of hyperparathyroidism in 1 case. In 2 cases the diagnosis was made through systematic radiological examination. Chest radiographs and neck and chest CT scans were done in all cases, and chest MRI was performed in 2 cases.

Table 1. Characteristics of patients with 'forgotten' goitre

Characteristic	Patient 1	Patient 2	Patient 3	Patient 4	Patient 5
Sex, age (years)	F, 32	M, 56	F, 49	F, 44	M, 50
Time since first thyroidectomy	8 years	6 years	6 months	3 years	4 years
Histological findings (first thyroidectomy)	Benign multinodular goitre	Benign multinodular goitre	Benign multinodular goitre	Benign multinodular goitre	Benign multinodular goitre
Symptoms	Dyspnoea, dysphonia, signs of hyperthyroidism	Incidental	Dyspnoea	Dyspnoea, dysphonia, chest pain	Incidental
Radiological findings					
Chest radiograph	Left tracheal deviation	Right tracheal deviation	Right tracheal deviation	Right tracheal deviation	Right tracheal deviation
Neck/chest CT scan	Right, 12 cm x 6.5 cm x 5.5 cm	Left, 8 cm x 5 cm x 4 cm	Left, 11 cm x 6.5 cm x 5.5 cm	Left, 10.6 cm x 7.6 cm x 5 cm	Left, 7 cm x 6.3 cm x 5.2 cm
Chest MRI	Not done	Not done	Not done	9.8 cm x 9.3 cm x 6.7 cm	6.7 cm x 6 cm x 5 cm
Hormonal findings	Hyperthyroidism	Euthyroidism	Euthyroidism	Euthyroidism	Euthyroidism
Surgical approach	Partial median sternotomy	Total median sternotomy	Partial median sternotomy	Total median sternotomy	Partial median sternotomy
Postoperative complications	Transitory left recurrent nerve paralysis	None	None	None	None
Histological findings	Benign multinodular goitre	Benign multinodular goitre	Benign multinodular goitre	Benign multinodular goitre	Benign multinodular goitre
Hospital stay (days)	10	5	6	7	5

F = female; M = male; CT = computed tomography; MRI = magnetic resonance imaging.

Standard chest radiographs (Fig. 1A) revealed a mass in the upper mediastinum with tracheal deviation or compression, and tracheal stenosis in 1 case.

The goitre extension was on the left side in 4 cases (Fig. 1A) and on the right side in 1 case.

Neck and chest CT scans (Fig. 1B and C) showed the masses to be situated in the upper mediastinum. The average size of the goitres was 9.7 cm x 6.4 cm x 5 cm.

Two patients had both CT and MRI scans (Fig. 2). On T1-weighted imaging the masses were hypo-intense and on T2-weighted imaging they were hyper-intense. Injection of gadolinium enhanced image quality.

Thyroid hormone levels were normal in 4 cases. One patient had hyperthyroidism.

All the patients were operated on via sternotomy (total in 2 cases and partial in 3 cases) (Fig. 3).

The findings on postoperative laryngoscopy were normal in 4 cases. One patient had a slight decrease in mobility of the left vocal cord. This transitory left recurrent nerve palsy was treated with phonatory rehabilitation sessions. The hospital stay was 5 - 10 days (mean 6.6 days).

In all cases, pathological examination of the goitre revealed multi-heteronodular thyroid

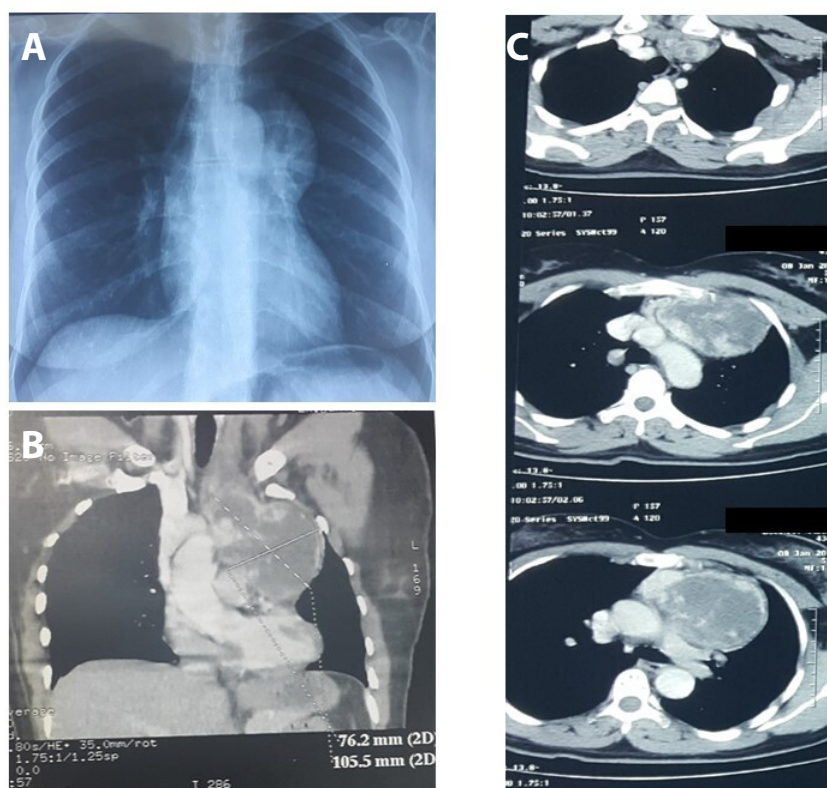


Fig. 1. (A) Chest radiograph showing a large well-defined opacity in the left hilar region. (B and C) Chest computed tomography scan showing a well-defined, slightly rounded mass of thyroid tissue that descends into the mediastinum down to the hilum.

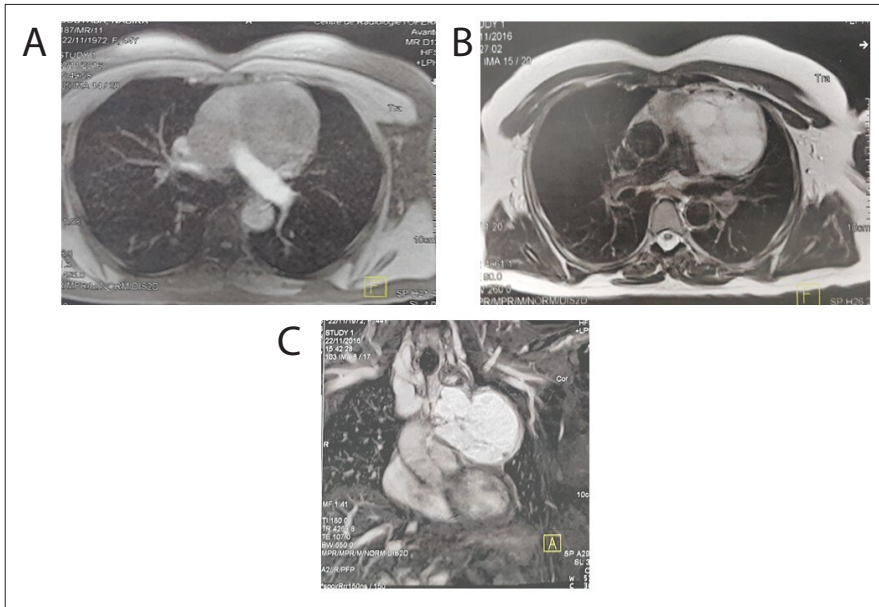


Fig. 2. Axial (A, B) and coronal (C) magnetic resonance imaging showing a mediastinal lesion with high T2 signal intensity.

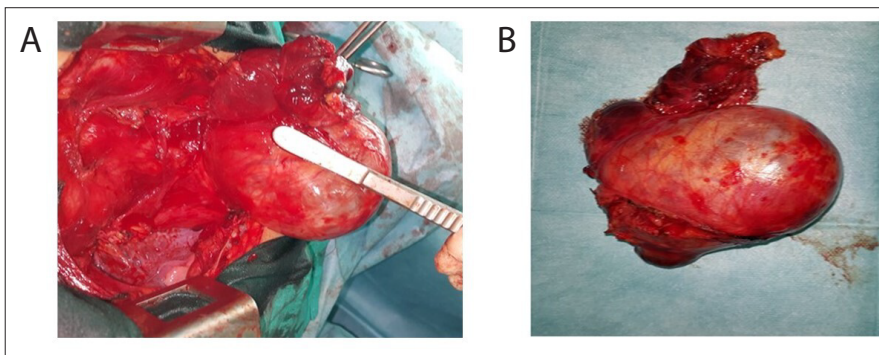


Fig. 3. Operative views. (A) Resection of a mediastinal forgotten goitre through a sternotomy. (B) The resected missed goitre.

hyperplasia without signs of malignancy. All the patients recovered uneventfully.

Discussion

'Forgotten' goitre or 'missed thyroid gland' is an extremely rare condition, defined as the presence of thyroid tissue as a mass in the mediastinum after total thyroidectomy.^[2] In the majority of cases, the 'forgotten' gland is the result of incomplete resection of plunging cervical goitre, but sometimes it is due to existence of intrathoracic autonomous ectopic goitre without parenchymal or vascular connections to the thyroid.^[2,3,6] The appellation 'forgotten' was used in 1992 by Massard *et al.*,^[2] referring to 'mediastinal masses of thyroid tissue diagnosed after subtotal thyroidectomy for substernal goiter that do not have visible connection to the cervical thyroid gland'.

Two situations are possible: either the retrosternal portion of the thyroid is connected to a cervical portion that was forgotten during total thyroidectomy (or not resected), or occasionally there is an autonomous goitre (estimated incidence 0.19%) located in the mediastinum without connections (parenchymatous or vascular) to the cervical part of the thyroid gland.^[2,7,8] An autonomous goitre is related to an abnormal embryonic progression of the thyroid gland. The ectopic thyroid tissue therefore results from the abnormal embryological migration of thyroid tissue, the anatomical communications between the neck and the chest (mediastinum), and the presence of pyramidal lobe, thyrothymic and Zuckerkandl remnants.^[9]

The incidence of FG is between 2% and 16% of retrosternal goitres in large series,^[3] and its

occurrence has usually been due to oversight rather than the autonomous ectopic forms. Our PubMed and Google Scholar literature search discovered ~40 cases (Table 2). According to Sackett *et al.*'s^[10] classification, remaining thyroid tissue can be missed after total thyroidectomy owing to impermanence of the link between thyroid tissue and the remnants in 20% of cases. Close attention must therefore be paid to preoperative imaging and intraoperative management during the first operation^[9] to avoid missing thyroid tissue.

The symptoms of FG are compression of mediastinal structures or signs of hyperthyroidism, so it should be strongly suspected when postoperative thyroid hormone (TSH) levels remain unchanged. Sometimes, however, the mediastinal mass is discovered incidentally in asymptomatic patients.^[1,3] The latent period between the first thyroidectomy and discovery of the FG is variable: 1 day in the Ismail *et al.*,^[11] report, 2 - 39 years in Massard *et al.*,^[2] and 1 month - 25 years in Courvoisier *et al.*^[12] In our case series the average period was 4.3 years.

Preoperative neck and chest CT scanning is essential to identify the exact size and location of the mass and any adhesions to mediastinal vessels, the trachea and the oesophagus. The CT scan also enables investigation of the characteristics of the mediastinal thyroid mass such as regular contour, presence of calcifications in the mass, high density and intense intake of contrast medium,^[1-3] and planning of the surgical approach to minimise operative complications. Coronal and sagittal planes in the chest MRI scan enable precise identification of all the adjacent organs, particularly the great vessels.^[1,3] Thyroid scintigraphy is useful in patients with a past history of thyroid resection. However, sometimes thyroid scintigraphy is negative because the thyroid tissue is positioned deep in the mediastinum or there is low absorption of the tracer photons by the sternum and other surrounding tissues.^[2,3]

Surgery is the best treatment for FG when possible. It confirms the diagnosis and relieves mediastinal compression. The choice of approach (cervicotomy, sternotomy, thoracotomy or a combined approach) is determined by the exact topography of the mass and its relationship with the mediastinal structures.^[1-3,13] The simple approach via repeat cervicotomy is probably preferable when

Table 2. Summary of cases of 'forgotten' goitre in the literature

Reference	Cases, N	Sex, age (years), range (mean)*	Symptoms	Time since first thyroidectomy, range (mean)*	Histological findings, first thyroidectomy	Surgical approach	Histological findings of the 'forgotten' mass
Calò <i>et al.</i> , ^[1] 2012	7	6 F, 1 M, 25 - 70 (56)	4 asymptomatic 2 mediastinal compression 1 persistent hyperthyroidism	-	NR	4 cervicotomy 3 partial sternotomy	No neoplasia
Massard <i>et al.</i> , ^[2] 1992	7	5 F, 2 M, 47 - 67 (56)	4 mediastinal compression 1 hypertyroidism 2 asymptomatic	2 - 39 years (16.1 years)	No neoplasia	1 cervicotomy 3 sternotomy 3 cervico-sternotomy	No neoplasia
Lucchini <i>et al.</i> , ^[4] 2014	4	3 F, 1 M, 46 - 56	4 asymptomatic	Mean 10 years	1 thyroid cancer metastasis in laterocervical nodes 3 nodular hyperplasia	1 cervicotomy 2 cervico-sternotomy 1 cervico-sternotomy and right posterolateral thoracotomy	1 follicular carcinoma 3 nodular hyperplasia
Sahbaz <i>et al.</i> , ^[6] 2013	2	F, 37 M, 72	2 persistent hyperthyroidism	4 weeks - 7 years	1 papillary cancer 1 Graves' disease	1 mini-sternotomy 1 sternotomy	1 papillary cancer 1 papillary carcinoma
Patel <i>et al.</i> , ^[8] 2016	1	F, 54	Incidental chest X-ray for routine surgery after 5 years	5 years	Hashimoto's thyroiditis	Trans-cervical	Nodular hyperplasia
Faroq <i>et al.</i> , ^[9] 2018	1	F, 58	Persistent hypertyroidism	~2 years (23 months)	Micropapillary carcinoma	Cervicotomy	Follicular lesion
Ismail <i>et al.</i> , ^[11] 2019	1	F, 57	Incidental imaging for other health condition	1 day	Multinodular goitre without malignancy	Cervicotomy	Multinodular goitre without malignancy
Courvoisier <i>et al.</i> , ^[12] 2015	11	8 F, 3 M, 34 - 71 (56.6)	NR	1 month - 25 years (10.6 years)	NR	4 cervicotomy 1 cervico-sternotomy 4 thoracotomy (only 9 operated on)	2 invasive thyroid cancer 7 benign goitre
Grigoletto <i>et al.</i> , ^[14] 1997	1	F, 59	Respiratory distress + cough	7 years	No neoplasia	Cervicotomy	No neoplasia
Khan <i>et al.</i> , ^[15] 2016	1	F, 59	Dyspnoea + stridor 25 years after thyroidectomy	25 years	Benign multinodular goitre	Partial median sternotomy	Benign multinodular goitre
Lee <i>et al.</i> , ^[16] 2005	1	F, 27	Incidental imaging for other health condition	35 months	Adenomatous hyperplasia	Sternotomy	Adenomatous hyperplasia
Casadei <i>et al.</i> , ^[17] 2002	1	M, 59	Mediastinal compression	3 months	Nodular hyperplasia with 7 mm follicular microcarcinoma	Sternotomy	Nodular hyperplasia

(continued)

Table 2. (continued) Summary of cases of 'forgotten' goitre in the literature

Reference	Cases, N	Sex, age (years), range (mean)*	Symptoms	Time since first thyroidectomy, range (mean)*	Histological findings, first thyroidectomy	Surgical approach	Histological findings of the 'forgotten' mass
Kesici <i>et al.</i> , ^[18] 2015	1	F, 49	Incidental after scintigraphy	NR	Papillary carcinoma	Partial median sternotomy	Nodular hyperplasia and chronic thyroiditis
Tsakiridis <i>et al.</i> , ^[19] 2016	1	M, 63	Dyspnoea + dysphagia	2 years	Multinodular goitre without malignancy	Cervico-sternotomy	Multinodular goitre without malignancy
Paczkowska <i>et al.</i> , ^[20] 2020	1	F, 88	Subclinical hyperthyroidism	16 years	Multinodular goitre	Radioactive iodine (20 mCi I-131) x 2 doses	No surgery
Kabiri <i>et al.</i> , 2021 (present study)	5	3 F, 2 M, 32 - 56 (46.2)	3 dyspnoea 2 incidental imaging for other health condition	6 months - 8 years (4.3 years)	5 benign multinodular goitre	Median sternotomy 3 partial 2 total	5 benign multinodular goitre

F = female; M = male; NR = not reported.

*Where applicable.

possible, because it is less traumatic with simple postoperative care, a short hospital stay, and a quick return to normal life and physical and professional activity.^[1,4,8,9,13,14] However, fibrotic cleavage planes after the first thyroidectomy, and deep mediastinal adhesions after a long delay (>2 years is usually considered a long delay in this context), make surgical resection unsafe and difficult.^[1,3,4] In this situation, sternotomy is the preferred procedure in view of the serious risk of mediastinal vascular injury and the possibility of a fatal outcome.^[2,3,6,15-18] This method is straightforward and safe, with good cosmetic results and a low rate of postoperative complications, similar to that of cervicotomy.

A right posterolateral thoracotomy is indicated in specific cases (right lateralised masses) according to the findings on preoperative imaging,^[3,4,12] offering good exposure and full control of the right mediastinum. Courvoisier *et al.*^[12] undertook resection of FG in 4 cases using the thoracotomy approach alone, but in one of the cases reported by Lucchini *et al.*,^[4] this approach was used to complement cervico-sternotomy.

Common complications of thyroid surgery were not reported in the literature reviewed, apart from a high rate of recurrent nerve palsy. Courvoisier *et al.*^[12] reported a high rate of nerve complications of 44% (4 cases of nerve palsy in 9 patients operated on). This high risk of recurrent nerve injury may be explained by repeat surgery, cervical fibrosis, and digital methods of extraction through a cervicotomy. The use of intraoperative recurrent laryngeal nerve monitoring and new haemostatic devices for dissection has significantly reduced the risk of recurrent nerve damage.^[3,4,19] The parathyroid glands are rarely exposed to surgical trauma, especially in the absence of a cervical approach.

Paczkowska *et al.*^[20] reported a case of FG in an 88-year-old woman, who was treated with radioactive iodine (20 mCi I-131) because of her age. A second dose of 20 mCi I-131 was necessary 1 year later. After 15 months the patient remained euthyroid and the goitre had decreased in volume.

Conclusion

FG is a rare condition. It should be considered in patients with previous thyroid surgery in the appropriate clinical context. It is possible to prevent FG by preoperative scanning and appropriate postoperative thyroid disease management. The best treatment for FG is still surgery.

Data availability. The datasets generated and analysed during the present study are available from the corresponding author (EHK) on reasonable request.

Declaration. None.

Acknowledgements. None.

Author contributions. EHK: conceived the study design, performed the operations, and reviewed the manuscript. MEH: collected data and drafted the manuscript. MB: collected data and analysed clinical information. MK: provided academic supervision and critically revised the manuscript.

Funding. None.

Conflicts of interest. None.

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Submitted 13 February 2023. Accepted 11 September 2024. Published 2 June 2025.