

Forgotten goiter

Our experience and a review of the literature



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Forgotten goiter. Our experience and review of the literature

"Forgotten" goiter is an extremely rare disease which is defined as a mediastinal thyroid mass found after total thyroidectomy. The authors report their experience of 7 cases of residual mediastinal goiter after total thyroidectomy for substernal goiter.

Six patients were female and one male with a median age of 56 years (range: 25-70 years). Four patients were asymptomatic, 2 patients showed signs of mediastinal compression, and one patient had persistent hyperthyroidism.

A sternal splitting incision associated with a cervical incision was required in 3 patients while an exclusively cervical incision was sufficient in 4 patients. Histology did not show neoplasia. Postoperative outcome was uncomplicated in 6 patients and in one patient a transient recurrent laryngeal nerve paralysis occurred.

In the majority of cases forgotten goiter is the consequence of the incomplete removal of a plunging goiter, although sometimes it may be attributed to a concomitant, unrecognized mediastinal goiter which is not connected to the thyroid. The residual goiter has the same clinical presentation as an ordinary intrathoracic goiter. Our experience confirms that surgical treatment of forgotten goiter is associated with only minimal morbidity although a sternal split is sometimes required. Forgotten goiter is a rare pathology which can be prevented if particular attention is paid to preoperative imaging and intraoperative management during the first operation. However surgical treatment for forgotten goiter, when performed in specialized centers, is associated with low morbidity

KEY WORDS: "Forgotten" goiter, Substernal goiter, Thyroid surgery.

Introduction

The first to use the term "forgotten goiter" ("goitre oublié") was Massard¹ when referring to "mediastinal masses of thyroid tissue diagnosed after subtotal bilateral thyroidectomy for substernal goiter, that do not have any visible connection to the cervical part of the thyroid gland". From a nosological point of view, this def-

inition is used for goiters that are difficult to classify since they are neither clearly cervico-mediastinal, plunging goiters nor an ectopic goiters.

In the literature the incidence rate of forgotten goiter ranges from 1.9%-16%^{2,3}. The pathology is most commonly found in women around 50 years old^{2,4,5}.

Since forgotten goiter is not rare and is the result of a technical error, we evaluated the causes of this condition and the measures that could be taken to reduce its incidence, based on our experience and a review of the literature.

Materials and methods

Between September 2002 and December 2010, 2512 patients underwent total thyroidectomy in the Department of Surgery at Monserrato Polyclinic, University Hospital,

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Cagliari. Two hundred and one of these patients (8%) had a cervicomediastinal goiter, and 7 (0.2%, 3.78% out of all substernal goiters), had forgotten goiter. The average age of these 7 patients was 56 years (range 25-70 years) and 6 were female.

The interval between their initial thyroidectomy and rein-tervention was on average 7 years (range: 1 month-25 years).

Symptoms of mediastinal syndrome in 2 cases, and persistent hyperthyroidism in one case led to diagnosis. In the other 4 cases forgotten goiter was an incidental finding. Diagnosis was based on thyroid hormone levels, thyroid ultrasound and scintigraphy, and CT scan of the neck and chest (Fig. 1). In one case magnetic resonance imaging (MRI) was also performed because CT scan was ambiguous.

According to the classification proposed by Di Matteo ⁶, the goiter was prevascular in 6 cases and retrovascular in one case.

Results

As regards the surgical approach, cervicotomy sufficed in 4 out of 7 cases (Fig. 2) and a partial sternotomy was used in the other 3 cases.

Six patients had an euthyroid multinodular goiter, and one had a toxic multinodular goiter. Histological examination did not show any evidence of neoplasia.

Only one procedure-related complication occurred; one patient had temporary paralysis of the right recurrent laryngeal nerve. Unfortunately this patient already had residual hypomobility of the left vocal cord because of her prior thyroid operation. The resulting reduction in respiratory function made it necessary to perform a tracheostomy. Three months later, due to recovery of vocal cord function, the tracheostomy was closed.

No cases of permanent or temporary hypoparathyroidism, permanent recurrent laryngeal nerve paralysis, or hemorrhage were observed.



Fig. 1: A CT scan showing residual thyroid parenchima in the antero-superior mediastinum.



Fig. 2: An hourglass-shaped goiter (intraoperative image).

The average length of hospital stay was 4 days for the patients who underwent sternotomy and 3 days for those who underwent simple cervicotomy.

Discussion

Forgotten goiter is usually the result of incomplete removal of a substernal goiter, but can sometimes be an unrecognized mediastinal goiter with no connection to the cervical thyroid ^{4,5}.

A more or less recent history of thyroidectomy for cervicomediastinal goiter and poor compliance with replacement therapy (Levothyroxin: LT4), should raise suspicion of forgotten goiter.

The length of time that passes between a patient's first thyroid operation and the diagnosis of forgotten goiter depends mainly on the clinical picture. Obviously the appearance of symptoms like those of hyperthyroidism leads to an earlier diagnosis of the condition than does slowly progressing compression of mediastinal structures.

The latent period is rather variable ⁷: in 7 cases described by Massard it ranges from 2 years to 39 years (1). Sanna et al report a case with a 25-year latent period (8) and Grigoletto et al one with a 7-year latency ⁹. In case series (5 cases) reported by Fattovich et al, the average latent period was 1.5 years. ² As mentioned above, in our case series the average latent period was 7 years with a very wide range. Often a diagnosis is made even if the forgotten goiter is clinically silent: a chest x-ray or other imaging exam reveals a mass in the neck or mediastinum ⁷ as an incidental finding. Less frequently the patient comes to the surgeon's observation because of a real mediastinal syndrome (in these cases the symptoms can range from cough to dyspnea and dysphagia)¹⁰. In our experience, the diagnosis of forgotten goiter was made because of mediastinal syndrome in 2 out of 7 cases, and because of persistent hyperthyroidism in one case while in the other 4 cases forgotten goiter was an incidental finding in asymptomatic patients.

Computed tomography (CT) scanning and MRI are important tools for diagnosing this condition since they make it possible to identify residual thyroid tissue (greater density and greater uptake of contrast agent) ⁷ (Fig. 1). However, in clinical practice, there are rarely indications for this kind of imaging when the first thyroid operation is performed. In our series, only one patient underwent MRI at the time of her prior surgery. For all the others we followed the traditional diagnostic pathway consisting of thyroid hormone levels, ultrasound and thyroid scintigraphy, and at times fine needle aspiration. CT scanning and MRI are essential for planning reintervention because they help to clearly define anatomical relationships and above all to assess the vascularization of the residual thyroid tissue (which is also useful for ruling out ectopic goiter). Tc 99m scintigraphy does not seem to be decisive since it does not show high uptake in cases of concomitant replacement therapy with LT4 ⁷. The nature of the mediastinal mass can be determined with CT-guided fine needle aspiration biopsy, even though this method is not often used ⁷. All our patients underwent ultrasound examination and CT scanning of the neck and chest before reintervention so that the volume of the residual thyroid tissue could be assessed and the best route of access could be planned based on the anatomical relationships revealed. As mentioned above, one patient also underwent MRI because CT scan was inconclusive.

In most cases, forgotten goiter is prevascular and this was also true in our series. In Massard's series ¹ histological examination showed that all the forgotten goiters were colloid-cystic goiters and no thyroid cancer was found. Our experience confirmed these results.

The finding of a mediastinal mass is an indication for surgery, especially if the patient is symptomatic. The treatment of forgotten goiter poses other problems. The choice of the method of access must be based on the way the operative field is exposed and on what major anatomical structures are adjacent to the thyroid mass.

Preoperative imaging and assessment of the vascularization of the mass are therefore of great importance. Reoperation for forgotten goiter is associated with the risk of damage to the parathyroid glands and the recurrent laryngeal nerves ¹¹. For this reason the methods of access described in the literature range from cervicotomy via a sternal split, to thoracotomy ¹².

In our series partial sternotomy (via a sternal split), was used in 3 out of 7 cases. It is a simple and safe method to use mainly for prevascular goiters, which is associated with a low rate of complications, good cosmetic results, and a length of hospital stay similar to that of patients who undergo simple cervicotomy (4 days vs. 3 days in our patients).

Since most of the patients were not young, it was important to consider comorbidities ⁸, most frequently hypertension, insulin-dependent diabetes mellitus, chronic obstructive pulmonary disease, and chronic obliterative arteriopathy (ASA III).

Because the incidence of forgotten goiter is not negligible and treatment is associated with complications, we believe it is essential to prevent the occurrence of this condition, which is almost always the result of underestimating foreseeable situations. In cases of cervicomediastinal goiter special care must be paid to maneuvers like luxation of the partially submerged goiter, since, though luxation without doubt useful in resolving a critical moment of the operation, there can be a catch; a thin strand of tissue can be torn during luxation and cause part of the thyroid parenchyma to be forgotten in the mediastinum. The same thing can happen with a multinodular thyroid gland formed like an hourglass or a bunch of grapes, especially if it is partially submerged (Figg. 2-3). On the other hand, keeping to a minimum diagnostic investigations the most common of which is an ultrasound examination of the neck, makes it easier for this error to occur. Having information provided by ultrasound imaging throughout the operation and at the same time performing a careful examination of the gland and the thyroid loggia after thyroidectomy can definitely be of assistance in determining whether there is residual parenchyma.

For the same reasons, when a reoperation for residual goiter is planned it is essential to perform a CT scan or MRI since intraoperatively it is difficult to ascertain visually or by palpation whether there is residual thyroid tissue.

Special care must be taken when using new devices for hemostasis and exeresis because, paradoxically, they can facilitate the development of this complication. Fibrous strands of tissue connecting the cervical part of the thyroid with the mediastinal part(s), are targets of hemostasis/resection. Recently only the careful exploration of

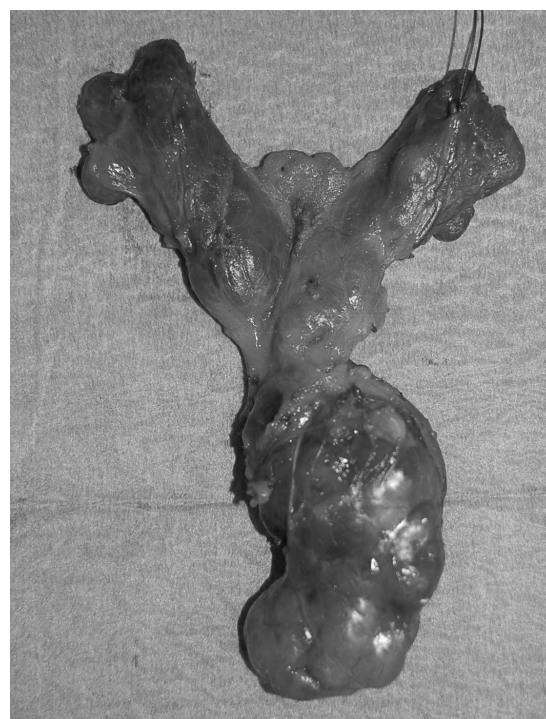


Fig. 3: An hourglass, shaped goiter (image of the surgical specimen).



Fig. 4: A thyroid gland like a bunch of grapes (visible on the left is a thin connection with the "forgotten" goiter accidentally cut with an ultrasonic scalpel).

the cervical loggia and the superior mediastinum prevented us from making this mistake (Fig. 4) .

Conclusions

If more care were taken by thyroid surgeons, the incidence of forgotten goiter, an uncommon condition that is not hard to diagnose, could be reduced to negligible proportions. Awareness of both the anatomical and technical causes of forgotten goiter is clearly important in this respect. Surgical management of the condition in specialized centers is associated with low morbidity.

Riassunto

Il gozzo dimenticato è una patologia estremamente rara definita come una massa mediastinica di natura tiroidea diagnosticata dopo tiroidectomia totale. Gli autori riportano la loro esperienza su 7 casi di gozzo dimenticato dopo tiroidectomia totale per gozzo cervico-mediastinico.

Sei su sette pazienti erano di sesso femminile, con età media di 56 anni (range: 25-70 anni); la sintomatologia che ha portato alla diagnosi è stata una sindrome mediastinica in due casi, un ipertiroidismo persistente in una paziente, mentre negli altri quattro la diagnosi è stata incidentale a seguito di altri accertamenti.

In 4 casi su 7 è stata sufficiente una cervicotomia, in tre è stata adottata una sternotomia parziale. In nessun caso è stata riscontrata una patologia neoplastica; in una

sola paziente abbiamo documentato una paresi transitoria del nervo laringeo inferiore, negli altri casi non è stata riscontrata alcuna complicanza.

Il gozzo dimenticato è in genere la conseguenza di una rimozione incompleta di un gozzo immerso, ma può talora essere attribuito ad un gozzo mediastinico non riconosciuto senza connessioni alla tiroide cervicale. Il gozzo dimenticato ha la stessa presentazione del gozzo mediastinico. La nostra esperienza conferma che il trattamento chirurgico può essere effettuato con una morbidità minima anche se uno split sternale è talora richiesto.

Il gozzo dimenticato è una situazione rara che può essere prevenuta con una particolare attenzione nella diagnostica preoperatoria e nella condotta intraoperatoria durante il primo intervento. L'intervento per gozzo "dimenticato" può essere effettuato con una bassa morbidità in centri specializzati.

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