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Management of Well-Differentiated Thyroglossal Remnant Thyroid Carcinoma: Time To Close the Debate? Report of Five New Cases and Proposal of a Definitive Algorithm for Treatment

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Background: Thyroid carcinoma is diagnosed in approximately 1% to 2% of thyroglossal duct remnants (TGDR). No definite agreement exists concerning the management of TGDR carcinoma, especially regarding the role of total thyroidectomy and postoperative adjuvant therapy. We report five new cases of TGDR carcinoma and review relevant articles in an attempt to clarify this issue.

Methods: We studied the demographic, clinical, tumor-related, treatment, pathologic, and outcome data on five patients treated at our institution for a TGDR carcinoma and compared the results with the four most important series published.

Results: All five patients were women, and diagnosis occurred after surgery in four. The sizes of the papillary tumors were 40, 38, 25, 23, and 15 mm (mean, 28.2 mm; range, 15–40 mm). Fine-needle aspiration biopsy of the TGDR had a low sensitivity (positive for one in four). A Sistrunk procedure was performed for resection of the TGDR in four patients, and plain TGDR resection was performed for the other patient. Three patients underwent repeat surgery; total thyroidectomy was performed in all cases, and cervical bilateral node dissection was performed in one case. With a median follow-up of 123.8 months (range, 8–284 months), all the patients are alive and free of disease, with no recurrences.

Conclusions: TGDR carcinoma is a rare malignant tumor that is usually diagnosed after surgery; papillary carcinoma is the most common type. The currently recommended treatment is a Sistrunk procedure, with a tendency to deferred total thyroidectomy in selected cases (similar criteria exist for papillary carcinoma of the thyroid gland). The prognosis is excellent, with a good long-term survival.

Key Words: Thyroglossal duct remnant—Papillary carcinoma—Thyroglossal carcinoma—Algorithm.

Thyroglossal duct remnants (TGDR), most typically cysts, are the most common congenital anomalies of the neck and represent >75% of childhood midline neck masses. Embryologically, the thyroid gland arises from a midline endodermal invagination

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of the foregut at the site of the future foramen cecum during the third week of fetal life. This invagination descends into the midline of the neck, traversing the mesoderm, which will eventually form the hyoid bone. By the seventh or eighth week of development, the thyroid reaches its normal position, the area below the thyroid cartilage, descending through the thyroglossal duct (TGD). For the 9th to 10th week of fetal life, the TGD disappears. If a remaining TGD fails to involute, it can persist as a cyst, a duct, or ectopic tissue, all of which are localized in the midline between the base of the tongue and the pyramidal lobe of the thyroid gland.[1] TGDRs are considered to be caused by mutations in genes responsible for the development of thyroid follicular cells (the thyroid transcription factors TITF1, TITF2, and PAX8 and the gene for the thyroid-stimulating hormone receptor [TSHR]).[2]

A histological analysis performed by Ellis and Van Nostrand[3] in 30 embryos, 200 adult larynges, and 20 thyroglossal cysts revealed that TGDRs fail to involute in approximately 7% of the population. Li Volsi et al.[4] reported that up to 62% of these may contain ectopic and functional thyroid tissue, thereby enabling the development of thyroid-related tumors. However, thyroid carcinoma arising in TGDR (TGDCa) is rare, and since the first report by Brentano[5] in 1911, only approximately 200 cases have been reported in the medical literature—most as single case reports. The first English-literature case report of a papillary TGDCa was written by Owen and Ingelby[6] in 1927. The true incidence of TGDCa is likely to be close to 1% of all TGDRs. Heshmati et al.[7] examined 741 TGD cysts excised at the Mayo Clinic and found an incidence of associated carcinoma of 1.62%, whereas Li Volsi et al.[8] found 6 (1.5%) in 377 and Doshi et al.[9] reported 14 (1.3%) among 1075.

The finding of TGDCa is a surprise for both the patient and the physician after adequate excision of the cyst, usually by means of a Sistrunk procedure (SP). The small number of patients in the series published, with only 3 articles in the literature reporting on >10 cases, makes it difficult to reach definite conclusions about diagnosis and treatment. The controversy in the treatment of this neoplasm mostly surrounds the optimal surgical management of the thyroid gland and the lymph node metastases (when present). We present a series of five cases diagnosed, treated, and followed up in our hospital over the last 21 years. We also review the recent relevant literature and propose a definitive algorithm for the rational treatment of this uncommon disease.

MATERIALS AND METHODS

A total of 511 patients who underwent surgical treatment for TGDR between 1983 and 2003 were identified from the prospective database of surgical patients maintained by the Endocrine Surgery Division at the Hospital Regional "Carlos Haya" in Malaga, Spain. Of these, five patients (1.21%) were found to have a TGDCa, accounting for 1.57% of the total number of thyroid cancers operated on during the same period. The medical records of the five patients were reviewed, and their pathologic specimens were reexamined. Criteria for inclusion in the group analyzed were clinical and operative evidence of a midline upper neck mass or cyst above the thyroid gland, surgical and gross anatomical diagnosis of TGDR tumor, and confirmed histological diagnosis of a malignant lesion. The information collected includes demographic, clinical, and pathological details of the tumor, as well as treatment and outcome data.

RESULTS

Table 1 expands on the preoperative, operative, pathologic, and postoperative follow-up of the series of patients. All the patients in our series were women with a mean age of 28.6 years (range, 22–51 years). None of the patients reported any predisposing factor such as previous neck irradiation. In three patients, the initial symptom was an enlarging mass in the neck, and in the other two an asymptomatic mass in the neck was discovered during a routine outpatient examination. The neck masses had been present for <6 months in four patients, and the other patient reported that the mass had been present for as long as 15 years before clinical evaluation. The masses were all firm, with two located in the suprahyoid area and the other three between the hyoid bone and the thyroid cartilage. The sizes of the papillary tumors were 40, 38, 25, 23, and 15 mm (mean, 28.2 mm; range, 15–40 mm).

All patients had normal results on thyroid function studies. Two patients had nuclear scanning of the thyroid bed, with normal uptake of the parenchyma and no areas of low or increased uptake near the TGDR. Three patients underwent preoperative

TABLE 1. *Profile of the five study patients with papillary thyroglossal duct remnant carcinoma in our series*

Variable	Patient 1	Patient 2	Patient 3	Patient 4	Patient 5
Age (y)	23	23	22	51	34
Sex	Female	Female	Female	Female	Female
Location	Midline upper thyroid	Midline upper thyroid	Suprahyoid	Suprahyoid	Midline upper thyroid
Evolution time of the cyst	15 y	6 mo	4 mo	2 mo	4 mo
Fine-needle aspiration biopsy	Not performed	Negative (false negative)	Positive (true positive)	Negative (false negative)	Negative (false negative)
Thyroglossal cyst diameter (mm)	40	38	25	23	15
Carcinoma diameter (mm)	Not available	Not available	20	2.5	6
Initial treatment	TGDR excision	SP	SP and TT	SP	SP
Secondary procedure	TT, SP, and CBND	TT	–	–	TT
Pathologic subtype of thyroglossal carcinoma	Papillary carcinoma (follicular variant)	Papillary carcinoma	Papillary carcinoma	Papillary carcinoma	Papillary carcinoma and delphian node (+)
Cyst wall invasion	Yes	No	No	No	Yes
Pathology of the thyroid	Multiple foci of papillary carcinoma	Normal	Normal	Previous NTT (colloid nodules)	Normal
Postoperative ablative I^{131}	Yes (×2)	Yes (×1)	Yes (×2)	Yes (×2)	Yes (×1)
l-T4 suppressive dose	Yes	Yes	Yes	Yes	Yes
Recurrence	No	No	No	No	No
Follow-up	24 y	12 y	8 y	7 y	8 mo
Actual status	Alive	Alive	Alive	Alive	Alive
Hypocalcemia/hoarseness	No	No	No	No	No

TGDR, thyroglossal duct remnant; SP, Sistrunk procedure; TT, total thyroidectomy; CBND, cervical bilateral neck dissection; NTT, nearly total thyroidectomy.

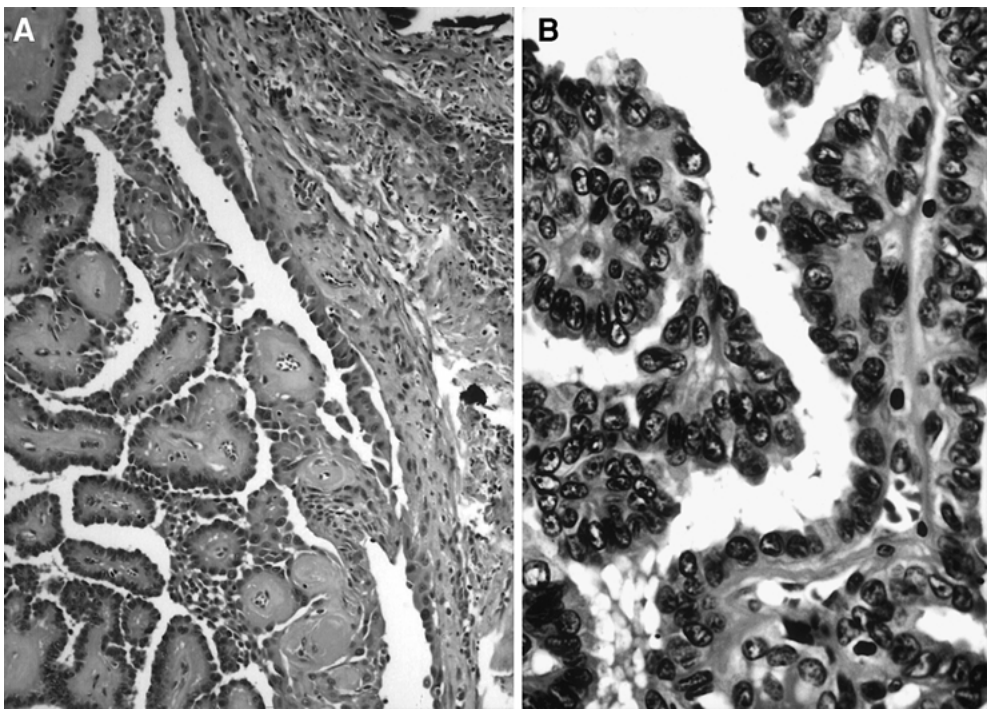

FIG. 1. Microscopic image of the focal infiltration of the wall of a thyroglossal duct remnant showing the central fibrovascular core with a line of cuboidal epithelial cells (stain, hematoxylin and eosin; original magnification, ×20 and ×40x for **A** and **B**, respectively).

ultrasonography (US). One patient had an anechoic, cystic mass in the midline, and the other two had heterogeneous, solid lesions containing hyperechoic and hypoechoic areas. Four patients underwent fine-needle aspiration biopsy (FNAB) that was not US guided: three aspirates showed a dark fluid

TABLE 2. *Summary of the largest case series reported of thyroglossal duct remnant carcinoma and comparison with our results*

Series	n	Sex (F/M)	% Carcinoma in TGDR	Period (y)	Preop cancer diagnosis
Heshmati (1997)[7]	12	6/6 (1:1)	12/740 (1.62%)	44	1/12 (8.33%)
Doshi (2001)[9]	14	8/6 (1.33:1)	14/1075 (1.3%)	24	3/14 (21.42%)
Patel (2002)[10,a]	62	37/25 (1.47:1)	5/37 (13.51%)	17	7/62 (11.29%)
Miccoli (2004)[18]	18	14/4 (3.5:1)	Not reported	26	10/18 (55.5%)
This study	5	5/0	5/412 (1.21%)	21	1/5 (20%)

and were reported as nodular and cystic colloid lesions (75% false-negative rate), and the other was informed correctly (the only patient who had a correct preoperative diagnosis; this resulted in a 25% true-positive rate for papillary carcinoma).

The SP was performed in all patients except patient 1, who only had the neck mass excised, and patient 3, who also had a total thyroidectomy (TT) because this was the patient whose preoperative biopsy showed papillary carcinoma. Treatment of patient 1 was completed at a second operation with TT, bilateral modified radical neck dissection, and SP because a lymph node was identified as positive for papillary cancer when the midline cervical mass was excised. Patients 2 and 5 underwent TT in a second procedure, and patient 4 had no further surgical treatment because a nearly total thyroidectomy had been performed a few years previously, with no evidence of tumor.

Histological examination of the tumor in the cyst showed a papillary carcinoma in all five cases, with cyst wall invasion in two cases (40%; Fig. 1). In four patients (including patient 4, who had previously undergone nearly total thyroidectomy), the thyroidectomy specimen showed either benign or normal tissue, and only one case (patient 1) had a multifocal papillary thyroid carcinoma and three lymph nodes involved in the right lateral compartment.

No patient had postoperative hoarseness or hypocalcemia, verified by the presence 1 month after surgery of normal postoperative blood levels of calcium and indirect laryngoscopy, which showed no alterations in vocal cord motility. After surgery, all five patients were treated with radioactive iodine at the ablative dose of 100 mCi and levothyroxine therapy at a suppressive dose, in accordance with our treatment protocol for differentiated thyroid carcinoma treated with TT. After a median follow-up of 123.8 months (range, 8–284 months), all five patients are alive with no evidence of recurrence, as shown by at least two negative whole-body scans with radioactive iodine and persistent low values of thyroglobulin.

DISCUSSION

Although TGDRs are the most common congenital midline neck masses, carcinoma is thought to be extremely rare, occurring in fewer than 1% of cases. Fewer than 200 cases have been reported since Brentano's first description[5] in 1911. Carcinoma arises slightly more often in females (female:male, 3:2; 100% women in our series), and the mean age of patients lies in the fourth decade of life, although the age ranges from 1 to 82 years and at least 31% of patients are < 10 years of age.[10,11] TGDR, which can be located anywhere along the embryological route of descent of the thyroid gland, are located as follows: 61% at the level of the hyoid bone or between the hyoid and the thyroid cartilage, 24% suprahyoid, 13% suprasternal, and 2% intralingual.

The clinical presentation of a TGDCa is often very similar to that of its benign counterpart; it is indistinguishable in terms of location, size, or consistency. Thus, in most cases a diagnosis of malignancy is not made until after surgery. Nevertheless, the carcinoma should be suspected in any remnant that is hard, fixed, or irregular or is associated with lymphadenopathy.[12] The differential diagnosis of central cervical cystic lesions involves many diseases, including thyroid diseases (benign colloid nodule, adenoma, papillary carcinoma, and cystic Hashimoto's thyroiditis in the pyramidal lobe), a delphian lymph node with cystic thyroid papillary carcinoma, a cystic parathyroid tumor and abscess, or a branchial cleft cyst located in the midline.[13,14] This variation explains why some authors recommend the systematic use of a US-guided FNAB in all these tumors.

Positive identification of TGDR is accomplished by demonstrating an epithelial lining of the duct and/or cyst and normal thyroid follicles in the wall. Consequently, a thyroid carcinoma is generally either of thyroid or squamous cell origin. Thyroid papillary carcinoma is the most common type (80%), followed by mixed papillary/follicular carcinoma (8%) and squamous cell carcinoma (6%). The other 6% includes Hürthle cell, follicular, and anaplastic carcinoma.[7,10] Widstrom et al.[15] described the

TABLE 2. *Continued*

Success of FNAB	Total thyroidectomy (%)	Presence of cancer in thyroid gland (%)	Lymph node disease (%)	Overall survival (%)
1/1 (100%)	75	33.3	25	100
3/5 (60%)	71.4	50	28.57	100
7/10 (70%)	48.4	26.7	11.29	100 at 5 y; 95.6 at 10 y
9/9 (100%)	100	33.3	16.6	100
1/4 (25%)	80	20	20	100

TGDR, thyroglossal duct remnant; Preop, preoperative; FNAB, fine-needle aspiration biopsy.
[a] This series is a compilation of all the cases of thyroglossal duct remnant carcinoma with adequate follow-up between 1984 and 2000. Patel et al. reported only five cases from the Memorial Sloan-Kettering Cancer Center.

definitive criteria for the diagnosis of primary TGDRCa: (1) the carcinoma should be in the wall of the TGDR, (2) the TGDRCa must be differentiated from a cystic lymph node metastasis by histological demonstration of a squamous or columnar epithelial lining and normal thyroid follicles in the wall of the TGDR, and (3) there should be no malignancy in the thyroid gland or any other possible primary site. This last criterion is the only point debated, because it excludes approximately 11% to 40% of all TGDRCa cases (patients who have a concomitant thyroid carcinoma). Patients with thyrogenic cancers may have a metastatic lesion from an occult primary lesion, and some physicians have suggested that a patent TGDR can be a route for metastatic involvement from the thyroid gland.[16,17]

Because TGDRs are usually associated with functioning thyroid tissue, either beside or inside the remnants, preoperative scintigraphy has little value and is generally considered unnecessary.[18] With US, thyroglossal cysts may be anechoic, hypoechoic, or complex heterogeneous lesions, and the cancer may appear as a mural lesion in the cyst, sometimes with microcalcification, or as a tumor invading the cyst wall.[19] On computed tomography and magnetic resonance imaging, the carcinoma can be seen as a solid nodule in the cyst (the most usual finding), an isolated calcification, an irregular margin, or a thickening in the wall of the cyst. A larger TGDRCa may present as a solid mass in the TGD or as a complex and sometimes invasive lesion in the midline of the neck.[20,21] When FNAB suggests TGDRCa, magnetic resonance imaging with gadolinium is performed to evaluate the neck for local invasion, cervical metastasis, and the presence of lesions within the thyroid gland itself.[22]

Bearing in mind the low prevalence of malignancy in TGDR (1%–2%), preoperative FNAB has been considered of questionable value because it is probably not cost-effective or appropriate, especially in children.[7,10,23] Nevertheless, Miccoli et al.[18] routinely perform FNABs and have reported an extremely high sensitivity (100%) and specificity (100%) in their recently published series of nine patients undergoing FNAB; this suggests that FNAB is a low-cost test if performed during a US scan but that it is highly operator dependent. In 2000, Yang et al.[24] found just 17 cases of TGDRCa diagnosed with FNAB in the English literature. The diagnostic accuracy was only 53% (9 of 17), and the false-negative rate was 47% (8 of 17) because of the hypocellularity of the specimens, which are diluted by the cystic fluid. Repeat FNAB (two or three times) of any residual mass after aspiration of the cyst may yield better results. In our experience, the success rate was very low (one of four; 25%), but we agree with Miccoli et al. and conclude that (1) FNAB is a relatively simply procedure, (2) FNAB should be considered in all adult patients with TGDR, and (3) any residual mass present once a TGD cyst has been emptied should be sampled, under US guidance, by either a clinician or, preferably, an interventional cytopathologist.

The definitive surgical management of TGDRCa remains controversial. The first step should always be to perform a wide complete excision by using the SP; a simple local excision of the TGDR is not now acceptable. The SP, originally described in 1928, considers the embryological development of the TGDR and includes en-bloc resection of the TGDR with the tract, the middle part of the hyoid bone (the body), and soft tissues along the course of the TGD to the level of the foramen cecum.[25] In the retrospective study published by Patel et al.,[10] from the Memorial Sloan-Kettering Cancer Center, univariate analysis of prognostic factors predictive of overall survival in patients with TGDCa revealed that the only significant predictor of outcome was the extent of surgery for TGDR. Patients who had simple excision fared significantly worse than those who had the SP, with 10-year overall survival rates of 75% and 100%, respectively.

The first question that always appears in the literature is whether the thyroid gland should be removed. Because normal thyroid remains are present in approximately two thirds of all TGDRs, most inves-

tigators believe that papillary carcinomas arise de novo from the TGDR rather than via metastatic spread from the thyroid gland, although a papillary carcinoma separate from the thyroid gland always raises the possibility of an occult primary carcinoma in the thyroid gland.[8] Other supporting evidence for the de novo theory is the fact that parafollicular cells are absent in ectopic thyroid tissue, and to date no case of medullary carcinoma of a TGDR has been published.[13] However, previously unsuspected thyroid involvement has been observed in 11% of TGDCa cases when the thyroid gland has been removed (which is not always done, so the real incidence may be underestimated). This involvement has increased in recent series[7,9,11,18] to 33% to 45% (20% in our series of five patients; Table 2) . Whatever the incidence, these figures lie within the .45% to 36% incidence of occult "incidental" thyroid carcinoma discovered in cadaveric thyroid tissue at autopsy.[26,27] Synchronous papillary carcinomas in TGDR and the thyroid gland represent multifocal tumors (ranging from .2 to 1.5 cm and mostly belonging to the microcarcinoma group) rather than metastatic spread. The effect of finding microscopic foci of well-differentiated thyroid carcinoma in an otherwise normal thyroid gland has been debated extensively and shown to have no effect on survival.[28,29] An absence of clinically recurrent disease after prolonged follow-up in patients not undergoing TT after SP is the norm in all the series. Thus, Patel et al.,[10] who used univariate analysis of prognostic factors to study overall survival in 62 previously reported cases, showed that the addition of TT to SP had no significant effect on outcome.

The SP is associated with a cure rate of 95% in papillary TGDRCa and a long-term survival ranging between 95% to 100% in all the series reported. Performance of TT to facilitate nuclear imaging studies and radioactive iodine ablation is not warranted. Only Miccoli et al.,[18] Heshmati et al.,[7] and Kennedy et al.[30] insist on recommending TT for a correct follow-up. The potential for serious complications from thyroidectomy (including a 3%–5% incidence of hypocalcemia and a 1%–2% incidence of recurrent laryngeal nerve injury) argues against further surgery. Thus, a TGDRCa must be considered a well-differentiated thyroid tissue cancer and must be treated as such with the same approach as for a well-differentiated carcinoma of the thyroid gland. Therefore, it is difficult to justify anything more than an SP for a patient with the following three criteria: (1) younger than 45 years and with no history of low-dose neck irradiation in childhood, (2) a clinically or radiologically normal thyroid gland, and (3) a small tumor (less than 1–1.5 cm or 4 cm, depending of the series) with an absence of histologically positive margins, cyst wall invasion, or metastatic lymph node spread.[31]

The incidence of cervical lymph node metastasis from a TGDRCa has been reported as 7% to 15%, far lower than that noted with papillary carcinoma of the thyroid gland, and death due to papillary TGDRCa is rare.[13,22,32,33] When cervical lymph node metastasis is present, the reasonable approach is to perform an SP, a TT, and appropriate level selective neck dissection with the same criteria as for the thyroid cancer, followed by postoperative radioactive iodine therapy. In the review by Patel et al.,[10] the univariate analysis revealed that the need for initial node dissection had no significant effect on outcome when compared with the group that did not require neck dissection. However, no definite conclusion can be drawn from the series because of the low number of patients, but the management is assumed to be similar to that for patients with papillary thyroid carcinoma.

The second question is what is the role of postoperative hormone suppression and/or ablation with radioactive iodine? Authors who defend TT always recommend postoperative radioactive iodine ablation and levothyroxine therapy at a suppressive dose with periodic whole-body scintigraphy during follow-up.[7,18] In patients treated with the SP, no data are available to clearly support the role of thyroid suppression. Nevertheless, most experts share the opinion that under these circumstances, the suppression of one possible stimulus to the thyroid tissue would be accomplished by keeping the thyroid-stimulating hormone between .1 and .5 mIU/L. Annual thyroid-stimulating hormone examination and a US scan of the thyroid gland should be all that is required for the patient's surveillance. Neither serum thyroglobulin measurement nor any studies with radioactive iodine are indicated.[22,34,35]

Squamous TGDRCa is a disease with a poorer prognosis, and only 11 cases with adequate follow-up have been described, with a disease-related mortality of 36.3% (4 cases). This carcinoma is usually more advanced at presentation, and local recurrence is more likely if excision is incomplete. Hannah[36] suggested that the best treatment for squamous TGDRCa is wide local excision with the SP and that neck dissection should be reserved for clinically involved nodes. Radiotherapy may be indicated in patients with larger lesions, positive margins, and extensive nodal disease.[36,37] In Fig. 2, we propose a definitive algorithm for the diagnosis, treatment, and follow-up of papillary thyroid carcinoma presenting in a TGDR.

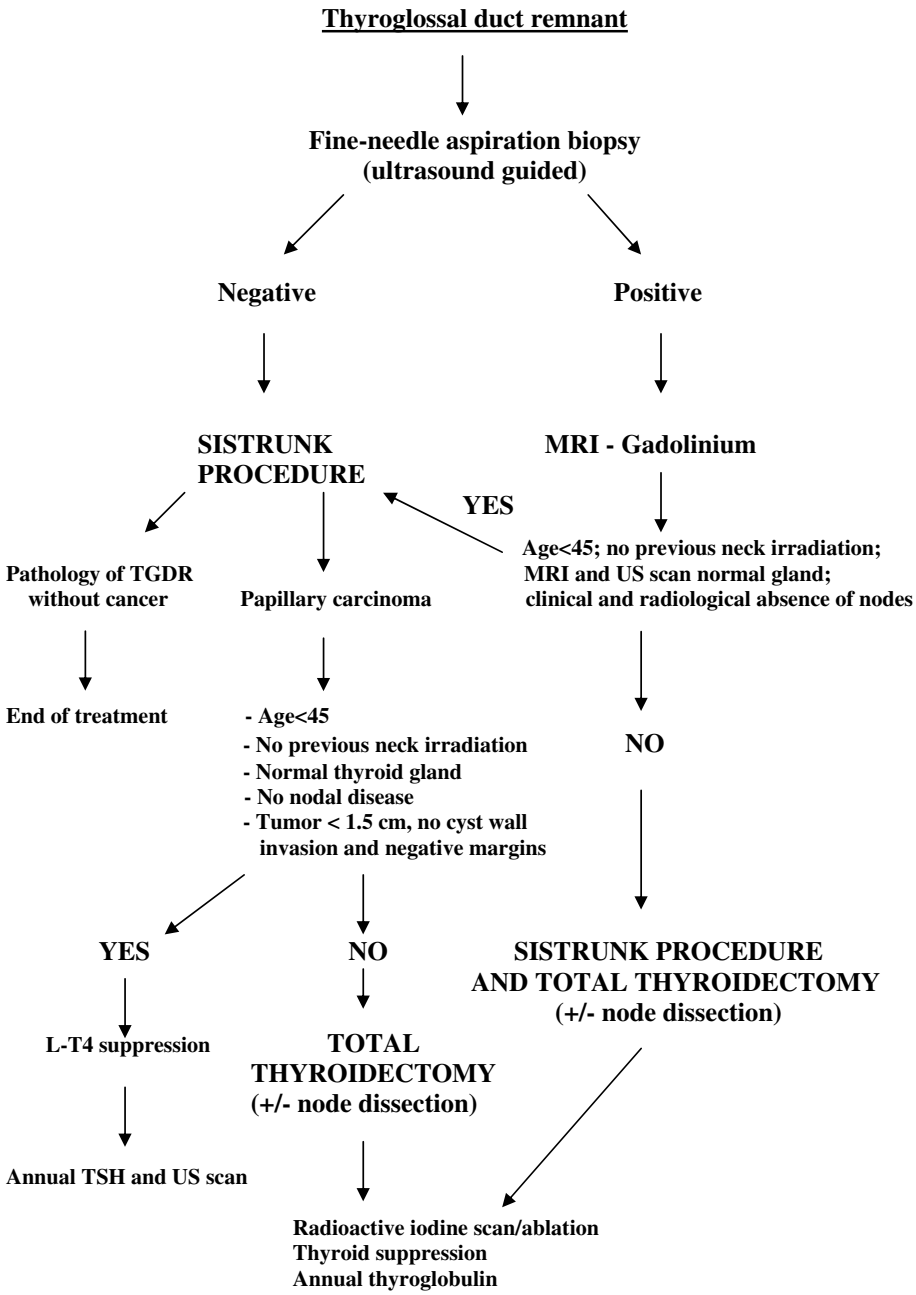

FIG. 2. Algorithm for the diagnosis, treatment, and follow-up of patients with papillary carcinoma in a thyroglossal duct remnant. MRI, magnetic resonance imaging; US, ultrasonography; TGDR, thyroglossal duct remnant; TSH, thyroid-stimulating hormone.

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