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Papillary carcinoma of hyoid

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ABSTRACT

INTRODUCTION: Thyroglosal duct cyst is a common anomaly with an incidence of 7% in adults, the rate of carcinoma in TGDC is 0.7–1.6%, and are extremely rare those originated in the hyoid bone.
PRESENTATION OF CASE: A 60 years old male patient, had a hard mass in the anterior neck. CT revealed a hyoid tumor. Hyoid bone resection was performed, the pathological report show a conventional papillary carcinoma in bone tissue. We rule out primary tumor in thyroid gland. Five years later, he developed a neck node recurrence. Total thyroidectomy and a selective left neck dissection (II–IV levels) was performed. He received radioiodine adjuvant treatment.
DISCUSSION: Hyoid cancer originates of a persistent thyroglosal duct remnants inside hyoid bone.
CONCLUSION: We propose to add a new subdivision to pathology derived from thyroglosal duct remnants). The diagnostic approach with ultrasound and CT are necessary. A primary in te hyoid gland mustang be discorded, and then the entire hyoid bone must be removed. Treatment of the thyroid gland and neck should be considered when there are significant risk factors of recurrence, similarly to thyroid cancer based on the risk assessment.

1. Introduction

The thyroid starts to develop at the 3–5 week embryo stage in the area that later becomes the foramen cecum, it descends in the midline through the muscle of the tongue, and then anterior to the hyoid bone and larynx (30% the tract mat be found posterior to hyoid), to override the fourth pharyngeal arch artery, and reaches its paratracheal position at the 7 week embryo stage, leaving the thyroglossal duct to obliterate and disappear by the 9–10. If obliteration and disappearance fail to occur, the remnants of the thyroglossal duct (TGD) are prone to become a thyroglossal duct cyst (TGDC) [1–6]. The TGDC had an incidence of 7% in adults, represents 70% of all congenital neck anomalies, and more than 50% contain ectopic functional thyroid tissue. It is more frequent in the first two decades of life [1,4,15]. The rate of carcinoma in TGDC is 0.7–1.6% [14], and most of times is a well differentiated papillary carcinoma, as thyroid carcinoma (PTC) less common is a follicular carcinoma [6].

The clinical presentation is a mass in the midline of the neck at hyoid bone. The epithelial lining of a TGDC ranges from squamous epithelium to pseudostratified ciliated columnar epithelium. Salivary gland tissue or thyroid gland tissue may be seen in the wall of the cyst [4,6].

Imaging studies displays a tumor at the level of hyoid bone. In Sonography it appears as well-defined cystic masses, with thin walls and late enhancement [7]. The FNA in TGDC had moderately sensitive [8]. The CT is very sensitive to evaluate bone, it is used to evaluate site of the mass, relationship with adjacent structures and characteristics, as thickness of walls, margins, internal septa, rim enhancement, internal density, and the presence or absence of the thyroid gland [9]

Surgical extirpation of TGDCs is recommended, because of the likelihood of recurrent infections and the rare possibility of malignancy [4]. The Sistrunk procedure has been a standard surgical procedure for the TGDC, the main aim is the complete removal of the cyst and the duct [2,4,10]. The recurrence rate is less 5–30%.

The Sistrunk operation is adequate for most patients with incidentally diagnosed TGDC carcinoma in the presence of a clinically and radiologically normal thyroid gland [4,13]. There are no reports on the surgical technique for hyoid.

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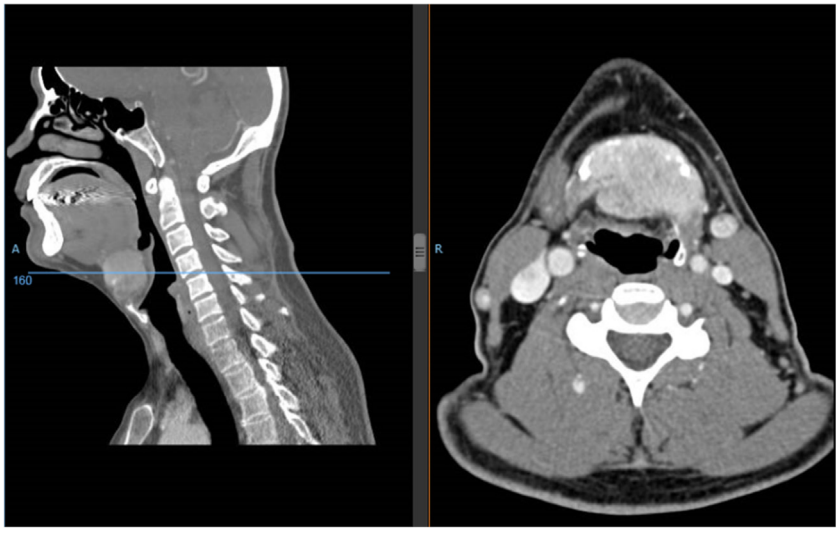

Fig. 1. CT scan. Large solid mass at the level of hyoid bone.

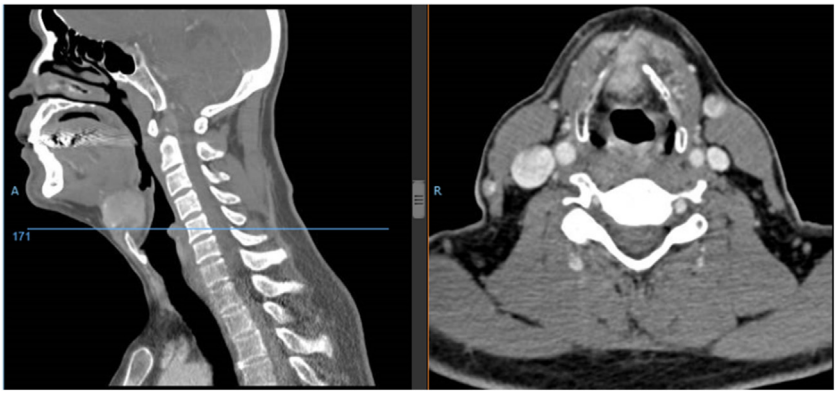

Fig. 2. CT scan. Thyroid cartilage extension.

2. Clinical case

60 years—old masculine patient, with history of smoking, presented an anterior midline neck mass that grew slowly and progressively over two years, associated with excessive salivation and hoarseness; Physical examination revealed a hard mass located in the anterior neck.

Thyroid functional tests, serum thyroxine (T4), free triiodothyronine (T3) and thyroid stimulating hormone (TSH), were within normal limits. A computed tomography scan (CT) revealed a large solid, heterogeneous mass, at the level of the hyoid bone of $55 \times 35 \times 39$ mm in size, which extended to the thyroid cartilage (Figs. 1 and 2).

We decided a surgical treatment. A hyoid bone resection was performed, finding a hyoid bone tumor of 7×4 cm. The pathological report revealed a suspected $4 \times 2 \times 1.1$ cm conventional papillary carcinoma with metastasis to the bone tissue and soft tissue extension (Figs. 3–5). US failed to show a primary in thyroid gland and nodal metastases. After multidisciplinary discussion, we suspected a primary papillary carcinoma of the thyroglosal duct remnant into de hyoid and decided to keep the patient under surveillance.

Five years later, physical examination revealed cervical lymph nodes; Neck ultrasound reported a suspected malignancy cervical lymph node (Fig. 6) and the fine needle aspiration (FNA) revealed a papillary carcinoma.

The patient underwent total thyroidectomy and left neck dissection of levels II – IV. The pathological evaluation revealed the thyroid gland with multinodular goiter without any neoplastic foci into thyroid gland and a metastatic papillary thyroid carcinoma was identified in 9 out of 17 lymph nodes (Figs. 7–9).

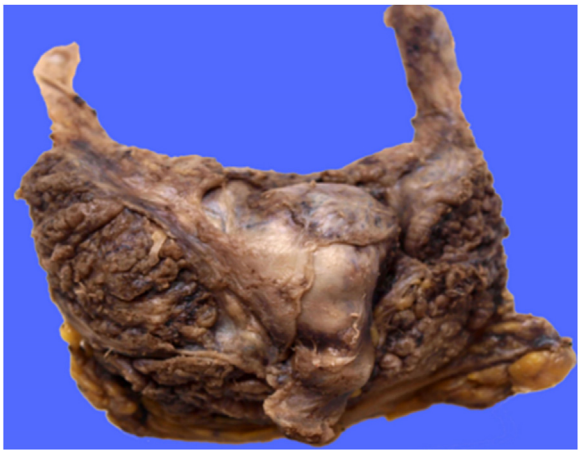

Fig. 3. Pathological report. Hyoid bone resection.

Postoperative radioactive iodine treatment and thyroid hormone suppression were recommended. At the present day, the patient was followed up with physical examination, thyroid functional tests and neck ultrasonography. The tumor had not recurred one year after the operation.

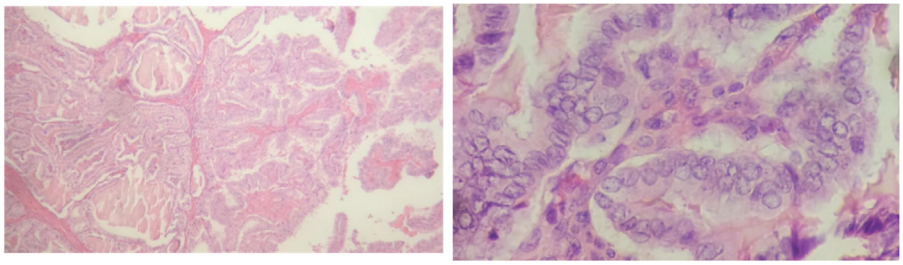

Fig. 4. Pathological report. Hematoxylin and eosin staining, was observed a malignancy forming papillae and follicles.

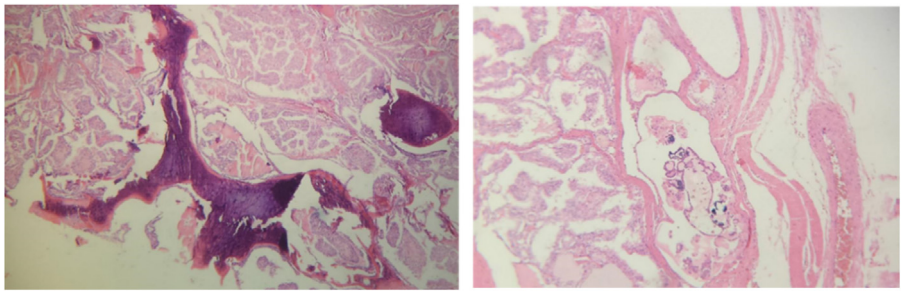

Fig. 5. Pathological report. Papillary carcinoma with bone infiltration and vascular permeation.

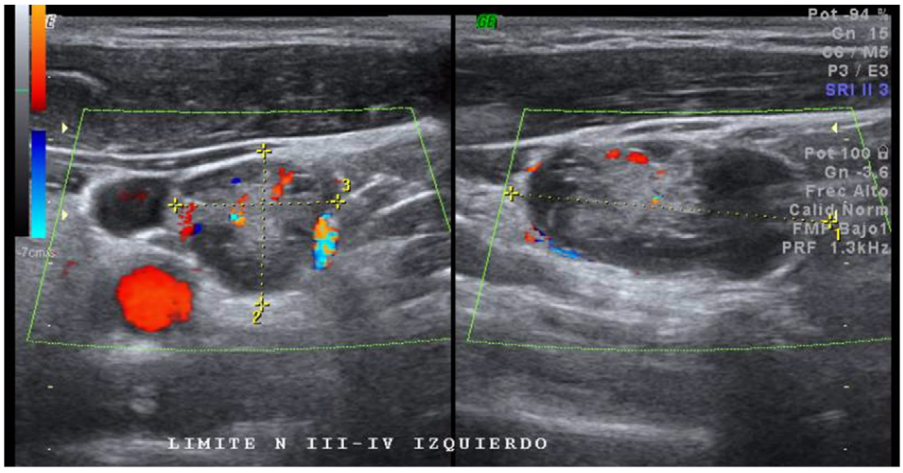

Fig. 6. Neck ultrasound. Lymph nodes with heterogenous echotexture, mixed (central/peripheral) blood vessels, located in left cervical levels IIA, IIB, III and IV.

3. Discussion

The rate of carcinoma in TGDC is 0.7–1.6% [14], of these, well differentiated papillary thyroid carcinomas (PTC) represents 66.7–95%, followed by follicular 16.7%, tall cell variant 11.1% and classic PTC with focal tall cell 5.6%; medullary carcinoma has not been reported in literature [6,14].

Of patients taken to thyroidectomy, 50% had incidental PTC with a size of 0.1–0.3 cm [6,14]. There is a pathological report of the thyroglossal duct penetrated the hyoid bone, result of a forward growth of the hyoid bone after first pushing the TGD outward, the shape of the bone adapts to the TGD and eventually completely engulfs it [2], this supports the diagnosis of hyoid cancer of this case report.

The locations of TGDC are classified into 4 subdivisions: (1) intralingual, (2) suprahyoid or submental, (3) thyrohyoid, and (4) suprasternal. We propose add a new subdivision "Hyoid".

The clinical presentation it's similar to a TGDC, with a mass in the midline of the neck at hyoid bone. The rest of TGDC are located just above or below the hyoid bone, but approximately one-third may present submentally or in lower cervical levels [4]. Less than 1% are located off the midline [1,4]. The epithelial lining of a TGDC ranges from squamous epithelium to pseudostratified ciliated columnar epithelium. Salivary gland tissue or thyroid gland tissue (22–46%) may be seen in the wall of the cyst [4,6].

Imaging studies revealed a tumor at the level of hyoid bone. Sonographic appearances in adults were identified: anechoic (28%), homogeneously hypoechoic with internal debris (18%), pseudosolid (28%), and heterogeneous (28%) [7]. The FNA in TGDC is moderately sensitive for a preoperative evaluation, with PPV of 69% [8]. But we believe that the main use of ultrasound is to assess the thyroid gland and cervical lymph node levels and guide a FNA. The CT is very sensitive to evaluate bone, It used to evaluate site of the mass, relationship to the midline, walls, margins, internal septa, rim enhancement, internal density, and the presence or absence of the thyroid gland [9]. We suggest that a patient with hyoid tumor must be evaluated with ultrasound and CT of the neck.

The Sistrunk procedure has been a standard surgical procedure for benign thyroglossal cyst, involves bloc cystectomy and central hyoidectomy, with tract excision up to the foramen cecum,

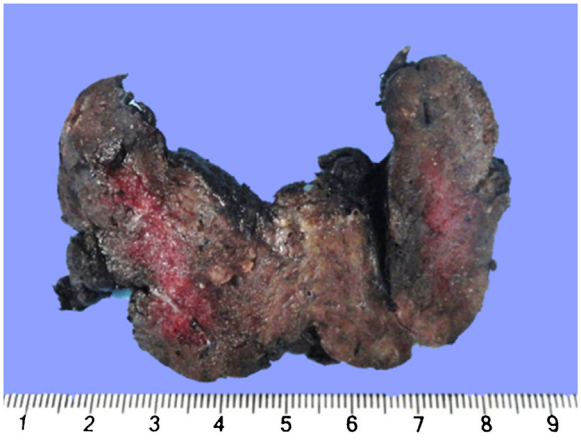

Fig. 7. Pathological evaluation. Total thyroidectomy and left neck dissection of levels II–IV. Thyroid gland without macroscopic tumor.

the main aim is the complete removal of the cyst and the duct [2,4,10,14]. The recurrence rate is less 10%, but are reports ranging 5–30%. The recurrence rate has been associate with an incorrect initial diagnosis in 50% of cases, previous infection in 15% of cases, unusual locations in 15% of cases, and lack of removal of tongue base muscle tissue and relative inexperience of the surgeon, both in 2% of cases [11,14].

The main complication of surgery for TGDC is infection (10–70%) [4]. Others complications are hematoma, blood transfusion, violation of the airway or tracheotomy, (hypoglossal) nerve paralysis, hypothyroidism, and death [12]. The posterior hyoid space is demarcated caudally by the inferior rim of the hyoid, cranially by the superior rim of the hyoid and thyrohyoid membrane, ventrally by the posterior surface of the hyoid, and dorsally by the thyrohyoid membrane facilitates complete resection and can prevent inadequate cyst resection in the area of the hyoid, reporting a recurrence rate of less than 2% [4].

The surgical management of TGDC carcinoma is based on the risk assessment, similar to well differentiated thyroid cancer, based on the revised 2015 American Thyroid Association (ATA) guidelines [15,16]. The Sistrunk operation is adequate for most patients with low risk with incidentally diagnosed TGDC (No local or distant metastases, all macroscopic tumor resected, not aggressive histology, no vascular invasion, without cervical metastasis, younger than 45 years, no radiation history and tumor less than 4 cm), the addition of total thyroidectomy did not have an impact on outcome, with a 95% cure rate [4,13,15]. In a patients with high risk are destined to an additional total thyroidectomy and postoperative radiactive iodine ablation therapy, the loco-regional control is a top priority [14,15].

There are no reports on the surgical technique for hyoid cancer but we consider should evaluate the absence of TGDC, and then remove the entire hyoid bone, the treatment of the thyroid gland and neck should be handled in the same way that a thyroid cancer; based on the risk assessment. In this case after a careful study, not primary tumor was demonstrated in the thyroid gland and cervical lymph nodes. The patient recurred in neck and the thyroidectomy was perform because at this time he had indications of radiative iodine ablation therapy.

This is the first report of a papillary carcinoma of hyoid, we offer a review of the embryological mechanisms leading to this pathology, as well as the diagnostic tools. We propose a new classification of thyroglosal duct remnants and appropriate surgical treatment for this patients.

4. Conclusions

The hyoid cancer originates of a persistent TGC result of a forward growth of the hyoid bone after first pushing the TGD outward, the shape of the bone adapts to the TGD and eventually completely

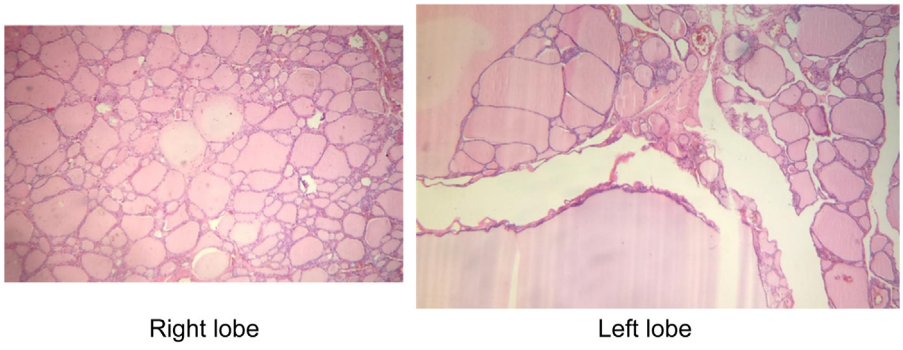

Fig. 8. Pathological evaluation. Total thyroidectomy and left neck dissection of levels II–IV. Thyroid gland with multinodular goiter.

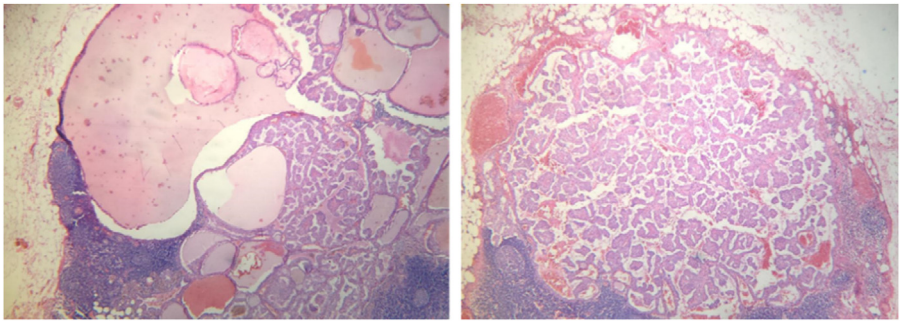

Fig. 9. Pathological evaluation. Total thyroidectomy and left neck dissection of levels II–IV. Lymph nodes with metastasis of papillary carcinoma.

engulfs it. We propose to add a new subdivision to the localization of TGDC (Hyoid). Histology and its frequency is similiar to the thyroid cancer. The diagnostic approach with ultrasound to assess the thyroid gland and cervical lymph node levels and lead a FNA and CT for evaluate bone. We consider should evaluate the absence of TGDC, and then remove the entire hyoid bone, the treatment of the thyroid gland and neck should be handled in the same way that a thyroid cancer; based on the risk assessment.

Conflict of interest

None of the authors have conflict of interest.

Sources of funding

None of the authors have sources of funding.

Ethical approval

Approval has been given.

Guarantor

Javier López-Gómez.

Consent

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

This work has been reported in line with the SCARE criteria [17].

Authors contribution

Dr. Javier López-Gómez—Design, data collection, data analysis, writing the paper. Dra. Ma. Alejandra Salazar-Álvarez—Data collection and writing the paper. Dr. Martin Granados-Garcia—Data analysis, data interpretación and writing the paper.

Research studies

In process.

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