

A case of branchial cyst with an ectopic thyroid papillary carcinoma



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Introduction

Cysts arising from the lateral neck are not uncommon. They more often represent benign lesions such as branchial cysts and lymphangiomas (also called cystic hygromas). Occasionally a cyst of the lateral cervical region is a malignant lesion metastatic from of a papillary thyroid carcinoma.

Ectopic thyroid tissue within a branchial cyst may give rise to an exceptional finding: a primary thyroid carcinoma. Only four of such cases have been described worldwide so far. With the patient recently seen in our institution we bring up to 5 the number internationally reported (1-4).

In these cases a thorough diagnostic work-up is needed in order to rule out a metastatic lesion from a primary located within the thyroid gland and even that may not be resolvable.

Case report

M.A.M. is a 29 year old female presenting with a history of a right lateral neck cyst first observed 5 years earlier. At that time the cyst was about 1.5 cm and surgical excision was unsuccessfully attempted by another surgeon. The past medical history of this lady was otherwise unremarkable and she denied any previous neck radiation exposure.

Riassunto

UNA CASO DI CISTI BRACHIALE CON CARCINOMA TIROIDEO PAPILLIFERO

Il caso presentato è il quinto della letteratura mondiale di carcinoma papillifero della tiroide insorto in una cisti branchiale.

Tale diagnosi comporta l'esclusione della natura secondaria della lesione e quindi l'esecuzione di una tiroidectomia totale ed uno studio istopatologico estremamente minuzioso ed accurato.

La conoscenza della embriologia delle lesioni cistiche del collo è fondamentale per definire i criteri diagnostici e le linee guida terapeutiche.

Parole chiave: Carcinoma, tiroideo papillifero ectopico, Cisti branchiale, chirurgia del collo.

Abstract

We report the fifth case worldwide described of thyroid papillary carcinoma arising in a branchial cyst. A metastatic lesion from occult primary thyroid carcinoma has to be ruled out. The embryology of this cervical lesion is also discussed in order to point out the diagnostic criteria and the therapeutic guidelines.

Key words: Ectopic thyroid papillary carcinoma, Branchial cyst, neck surgery.

Physical exam only showed a 3 cm lateral right neck scar overlying a roughly 5 cm, egg-shaped, elastic, indolent, mobile mass. No skin changes were present.

She was referred to us because of the progressive growth shown by her cyst throughout the 5 year. Not only documented the imaging a now 6 cm lesion but also its cystic nature and its close relation with the anterior and lateral aspect of the right internal jugular vein. The thyroid gland was normal.

The patient underwent excision of what grossly appeared a benign, clear fluid-filled lesion resting on the right jugular vein, with no signs of invasion.

Pathology was consistent with a multiloculated, serous filled, 4 cm branchial cyst containing a 1 cm yellow ve-



Fig. 1: Operatory field: The lateral cyst is almost totally isolated.



Fig. 2: The cyst.

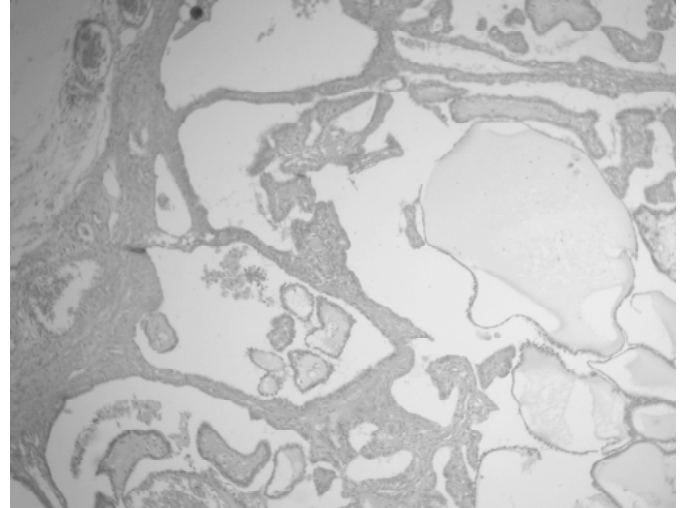


Fig. 3: The cyst wall is formed by fibrous tissue with lining by monostratified epithelial cells. A focal intracystic papillary carcinoma is shown (EE x 100).

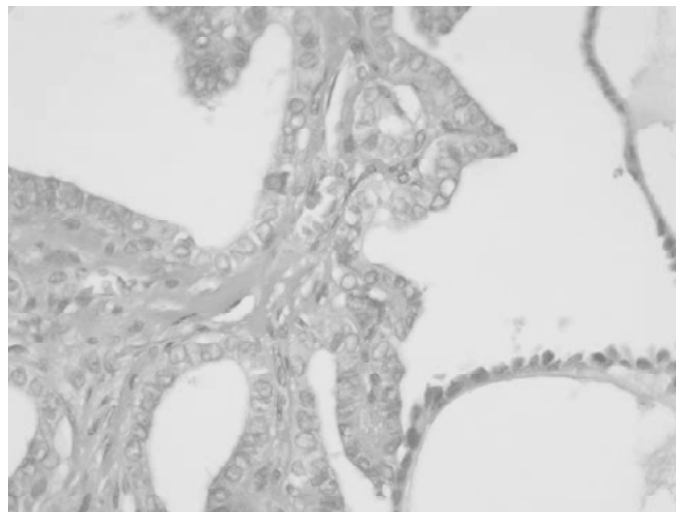


Fig. 4: High power field of papillary carcinoma with characteristic nuclear changes (EE x 400).

getation. A well differentiated papillary thyroid carcinoma was within it. Invasion of the cyst wall was absent. A single lymph node included in the specimen was tumor free.

Although physical exam and thyroid US were repeated and CT and MRI of the neck obtained before surgery were reviewed, no evidence of thyroid gland tumor was found.

Total thyroidectomy appeared ineluctable and after discussing with the patient the need for ruling out a primary gland tumor the operation was performed.

Pathology did not show any primary thyroid gland tumor and a whole body ¹³¹I scan done weeks after the operation failed to show any metastatic disease.

Discussion

In order to understand how ectopic thyroid tissue may be found inside a branchial cyst we must revisit the embryology of this gland.

The thyroid gland originates from the primitive alimentary tract and is predominantly of endodermal origin. It arises as a midline diverticulum from the floor of the pharynx in the region of the foramen cecum at about the third gestational week and becomes recognizable about 1 month after conception. The main body of the thyroid descends into the neck from its origin and migrates caudally. It then develops into a bilobated solid organ. With the resorption of the thyroglossal duct, the origi-

nal attachment to the buccal cavity at the foramen cecum, around the sixth week of gestation the developing gland reaches its final location.

According to Weller and more recently Hoyes, Kershaw and Williams follicular cells from the fourth branchial pouch contribute to the development of the thyroid gland lobes (5-7).

A failed obliteration of 2nd, 3rd and 4th branchial pouch was thought caused to be the etiology of branchial cysts. This theory has been abandoned and it is now believed that epithelial cell inclusions from the upper airway and GI tract enter the lymphatics and reach the lateral neck lymph nodes. This colonization would promote a "cystic degeneration" of lymph nodes (8-9).

It has been postulated that similar changes might occur if aberrant thyroid tissue colonizes the lateral neck lymph node as a result of an "embryologic accident".

Nevertheless no evidence has been found that such "accident" may actually occur and trigger a cystic degeneration of lymph nodes.

On the other hand it has to be kept in mind that an enlarged lateral neck lymph node might represent a metastasis of a small or occult thyroid gland primary tumor. Then the metastatic lymph node may undergo massive necrotic degeneration providing it with "cystic" features. Therefore in the differential diagnosis of a lateral neck cystic lesion we have to include benign lesions like the branchial cyst and the lymphangiomas but also metastasis from head-neck tumor including the thyroid gland. The evaluation of a patient with a cystic lesion of the lateral neck relies on those tools used for other neck masses. The FNAC most times allows to diagnose neoplastic lesions. Furthermore if a yellow fluid is retrieved during the FNA the likelihood of a branchial cyst is high. If a brown, chocolate-like fluid fills the "cyst" a metastatic lymph node is the most likely condition (10-12). Evidence of cholesterol crystals in the cyst content suggest a branchial etiology (1), whereas the presence of macrophage is not specific being these elements found in both the branchial (10) and the neoplastic, metastatic lymph node (13). Thyroglobulin can also be found (1-2).

CT scan is not helpful in the differentiating a branchial cyst from a metastatic lymph node with necrotic changes, although a visible intracystic vegetation may suggest the latter (10).

The result of pathology study may show thyroglobulin in the metastatic node and squamous or columnar epithelial lining in the branchial cyst.

Finally, in order to rule out a primary thyroid gland tumor, serial microscopic examinations on very thin sections have to encompass the whole gland. Only by a thorough exam small papillary carcinomas can be detected, as shown by Li Volsi (13). In her report a papillary thyroid carcinoma smaller than 1 mm was found in a patient who underwent excision of a metastatic neck lymph node 9 years earlier. In this case the primary thyroid tumor was not detected by any preoperative test or standard pathology sections.

Conclusion

A papillary thyroid carcinoma found within a cyst of the lateral neck represents a diagnostic and therapeutic dilemma.

The finding of such lesion within a branchial cyst does not allow to consider it as an aberrant thyroid excluding a primary papillary thyroid tumor. In our opinion the lack of evidence suggesting a primary papillary tumor mandate a total thyroidectomy and a very thorough examination of the gland.

The absence of a primary tumor after performing the thyroidectomy leaves the surgeon with the question of whether to add a modified radical neck dissection or not.

We recommend the neck dissection in the instance that the lateral neck cyst indeed reveals a metastatic nature with degenerative "cystic" changes.

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Commento

Commentary

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La principale insidia della chirurgia è forse rappresentata dalla capacità delle lesioni neoplastiche di camuffarsi sotto le spoglie più innocenti, e non sempre anche le più raffinate metodiche di imaging sono in grado di dirimere i dubbi preoperatori.

A volte, come in questo rarissimo caso, il dubbio si presenta al momento della diagnosi istologica, e nonostante un accurato studio preoperatorio, rappresenta l'avvio ad ulteriori indagini. Cappellani e coll., di fronte ad un carcinoma tiroideo ectopico, hanno affrontato con molta attenzione il problema anche embriologico (1-3) ma soprattutto clinico, e dopo una revisione accurata della letteratura e della problematica istologica ed oncologica, mostrano il modo a mio avviso più corretto di affrontare tale problematica e l'approccio diagnostico-chirurgico "ideale" (4).

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