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 resolutive.

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.

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-
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 ce-
sum, 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 pou-
ch was thought caused to be the etiology of branchial
cysts. This theory has been abandoned and it is now
believed that epithelial cell invasions from the upper
airway and GI tract enter the lymphatics and reach the
lateral neck lymph nodes. This colonization would pro-
mote 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 “accid-
ent” may actually occur and trigger a cystic degenera-
tion of lymph nodes.
On the other hand it has to be kept in mind that an
enlarged lateral neck lymph node might represent a meta-
stasis 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 meta-
stasis from head-neck tumor including the thyroid gland.
The evaluation of a patient with a cystic lesion of the
lateral neck relays on those tools used for other neck
masses. The FNAC most times allows to diagnose neo-
plastic 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 meta-
static lymph node is the most likely condition (10-12).
Evidence of cholesterol crystals in the cyst content sug-
gest a branchial etiology (1), whereas the presence of ma-
crophage 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 chan-
ges, although a visible intracystic vegetation may sugge-
t the latter (10).
The result of pathology study may show thyroglobulin
in the metastatic node and squamous or columnar epi-
thelial 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 small-
er than 1 mm was found in a patient who underwent exi-
sion 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 exa-
mination 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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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).

Bibliografia


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